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Developmental apraxia of speech:
deficits in phonetic planning and motor programming

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The illustration used on the front cover was realized by taking the utterance 'he ze schiet weer' spoken by a child with developmental apraxia of speech. Subsequently, this utterance was digitally manipulated by Louis ten Bosch resulting in the figure shown on the front. *[After an AD conversion of the utterance with a sampling frequency of 20 kHz, the spectrogram was calculated using a Hanning window of length 1024 samples. After computing the FFT, negative frequencies have been removed, and for each frame the logarithm of the absolute value of the resulting components were used. The frame shift was 341 samples. Next, the resulting frames were interpreted as a matrix, from which an augmented point-symmetric matrix has been constructed by gluing three appropriately mirrored copies to the original matrix. In the final step, the values in the augmented matrix have been interpreted as height values and a contour plot was devised on the basis of this height interpretation. The resulting figure has been chosen after aesthetically motivated manipulation of the corresponding colour map].* The back cover shows the utterances of the title and author's name spoken by the author, which are, after recording and digitizing, displayed as oscillogram waveforms.

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Developmental apraxia of speech: deficits in phonetic planning and motor programming

Een wetenschappelijke proeve op het
gebied van de Medische Wetenschappen

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Lian Nijland

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CHAPTER ONE

Introduction and Thesis Outline

INTRODUCTION

Speech is very important in our daily communications. Most of us take this ability to communicate for granted. We think of something to say, open our mouths, and the words come out. Although it may appear to be simple, producing intelligible speech is not that obvious for everybody. Children with Developmental Apraxia of Speech (DAS) suffer from a speech disorder that interferes with the ability to produce intelligible speech. Their speech is characterized by a large number of consonantal errors, (contextual-based) substitutions and omissions, without clear indications of an identifiable language problem (as is the case in children with a specific language impairment). Furthermore, inconsistency in errors and groping (a searching articulatory behavior) are typical for DAS (Hall, Jordan, & Robin, 1993). Especially this inconsistency makes it hard to understand the speech of children with DAS. For example, a word like 'speech' might first be pronounced as 'steetch', but a following attempt might lead to 'peep' or 'street', and also the correct production of 'speech' might occur. This inconsistency in repeated productions differentiates children with DAS from children with other articulation disorders, like children with dysarthria (a clearly neurologically based speech disorder) for instance, who show consistent error patterns and are therefore easier to understand. Prevalence estimates of DAS varies from 1-2 children per thousand (Shriberg, Aram, & Kwiatkowski, 1997b) up to 1% (Yoss & Darley, 1974). Another aspect of DAS is that it is highly resistant to classical articulation therapy procedures. Even when children with DAS receive many hours of speech therapy each week, they make little progress in learning to produce intelligible speech. In the literature, different views about the prognosis of DAS are reported and objective data are lacking. A number of studies mention spontaneous improvement in some children with DAS (especially in the mild cases) and others report disorders in adults ranging from 'minimal signs' to unintelligibility. However, most researchers suggest that, as adults, those individuals that were diagnosed with DAS may well become functional oral communicators, but that a 'normal' outcome is unrealistic. Upon close examination, subtle problems may become evident (for an overview see Hall et al., 1993). The intelligibility of their speech, due to the inconsistency in errors, and the resistance to therapy make this a very intriguing group for scientific research.

The diagnosis of DAS is fraught with controversy and concerns its definition, terminology, symptomatology, as well as its diagnostic features. Even the very existence of developmental apraxia of speech as a diagnostic entity is disputed. Although the existence of a group of children that demonstrate multiple articulation errors, effortful speech, and slow progress in remediation is rarely questioned, the nature, diagnosis, etiology, prognosis, and remediation of this disorder have been subject of debate for a century (Hall, 1992). In 1989, Geert Thoonen took on a research project in our center (the department of Pediatric Neurology of the University Medical Center Nijmegen) aimed at investigating the defining speech characteristics in children with DAS (Thoonen, 1998). The results of his research contributed to the characterization of children with DAS. His thesis provided a thorough description of objective and quantitative speech characteristics of children with DAS, on the basis of which appropriate diagnostic assessment procedures could be developed. Thoonen and colleagues adopted a standardized analysis procedure of transcriptions and diadochokinetic tasks (Thoonen, Maassen, Gabreëls, Schreuder,

& De Swart, 1997). Based on these results, they hypothesized that the underlying deficit¹ in DAS emerges during the phonological encoding stage and possibly during the transition from a phonological code into speech gestures. However, direct evidence for this assumption was lacking. The issue of the underlying deficit of DAS is addressed in the present thesis. We took up the line of research where it was left off by Thoonen et al. (1997) and conducted a more hypothesis-driven evaluation to try and establish the nature of the underlying deficits in DAS. For this purpose, we chose phonemically correct productions in children with DAS instead of patterns of errors as our focus. Before elaborating on the research topics of the present thesis, I will first provide an outline of the literature on the diversity in clinical symptoms and diagnostic classifications and the main theoretical views on the underlying deficits of DAS.

Symptoms and diagnostic issues

There is debate among researchers about the exact speech symptoms of DAS as well as the accompanying non-speech characteristics. Guyette and Diedrich (1981) noted that there are no pathognomonic (i.e., differential diagnostic) features to diagnose DAS and to differentiate DAS from other speech output disorders. Besides the characteristics mentioned above, also errors and distortions in vowel productions have been reported (Pollock & Hall, 1991; Walton & Pollock, 1993), as well as inappropriate stress patterns (Shriberg, Aram, & Kwiatkowski, 1997a). Non-speech oral motor actions and movements, like coughing, chewing, swallowing, and (pretending) licking an ice cream or blowing, do not necessarily cause difficulties, which differentiates DAS from oral apraxia. In addition, various studies reported additional problems in children with DAS, for instance, language or language-related problems (Groenen, Maassen, Crul, & Thoonen, 1996; Guyette & Diedrich, 1981; Hodge, 1994; Hoit-Dalgaard, Murry, & Kopp, 1983; Marion, Sussman, & Marquardt, 1993; McCabe, Rosenthal, & McLeod, 1998). Moreover, production as well as perception errors have been mentioned. A study by Hoit-Dalgaard et al. (1983), for example, shows that apraxic subjects demonstrate problems in both the production and the perception of the voicing features of phonemes. A similar relation between production and perception problems was found in children with DAS with respect to the feature ‘place-of-articulation’ (Groenen et al., 1996) in rhyming abilities (Marion et al., 1993). Children with DAS also often demonstrate ‘soft’ neurological signs, such as clumsiness and motor coordination problems (Guyette & Diedrich, 1981; Ozanne, 1995; Pollock & Hall, 1991; Robin, 1992).

More recent discussions concern the search for the ‘diagnostic marker’ for DAS. Shriberg et al. (1997a) suggested inappropriate stress as a diagnostic marker, which might be applicable to a subtype in approximately 50% of the children diagnosed with DAS. Some researchers discussed the possibility of a more general inability underlying DAS (Davis, Jakielski, & Marquardt, 1998; Velleman & Strand, 1994). The study by Thoonen, Maassen, Gabreëls, and Schreuder (1999) yielded a measure for degree of involvement of DAS based on the Maximum Repetition Rate (MRR), especially in trisyllabic repetitions. In contrast, involvement of dysarthria can be assessed on the basis of monosyllabic repetition rate. However, the above-mentioned study furthermore

¹ Note that the ‘underlying deficit’ refers to a deficit in a cognitive model of speech production; this should not be confused with a deficit in the neuroanatomic architecture.

showed that all children experiencing speech problems go through a stage in which they display characteristics of DAS to some extent. This underlined the importance of both monosyllabic and trisyllabic MRR as diagnostic criteria.

Despite the dispute about pathognomic features, there is more or less agreement about a set of more central or core diagnostic symptoms of DAS. These comprise a high number of consonant errors, especially substitution in place of articulation, inconsistency in repeated productions, difficulty in sequencing phonemes, especially in diadochokinetic tasks, groping, and resistance to therapy (also see Davis et al., 1998; Hall et al., 1993; Thoonen, 1998). Setting aside the ongoing debate, we adopted the selection criteria as proposed by Thoonen et al. (Thoonen, Maassen, Wit, Gabreëls, & Schreuder, 1996) and applied ourselves to the investigation of the underlying deficit in DAS with the aim to provide useful information that could contribute to a better understanding of the disorder. (Note that the Thoonen et al. criteria were developed for Dutch children. Validation of diagnostic criteria across languages has yet to be performed).

Clarification of the underlying deficit

In neuropsychological or neurolinguistic approaches a diversity of views exists also with regard to the underlying deficit. Explanations for DAS range from a disturbance localized at the level of phonological representation, the phonological encoding process, the generation of a phonetic plan, to the motor programming and execution levels (Ballard, Granier, & Robin, 2000; Dodd & McCormack, 1995; Hall et al., 1993; McNeil, Robin, & Schmidt, 1997; Ozanne, 1995; Shriberg & Kwiatkowski, 1982; Van der Merwe, 1997). In the following section, I will give an overview of the levels of speech production at which deficits might occur in children with DAS, and describe speech symptoms that might arise from a deficit at each level. This formed the basis for the studies of the present thesis. Subsequently, I will give a brief introduction to the experiments that constituted the body of my research. These experiments aimed to test in a more explicit manner the possible involvement of the successive levels of speech production in the underlying deficit(s) of DAS.

SPEECH PRODUCTION MODEL

Figure 1 shows a model of speech production in which the levels of speech production that might show deficits in children with DAS are shown. This model is based on the latest speech production model developed by Levelt (Levelt, Roelofs, & Meyer, 1999), which was founded on earlier models of Levelt (Levelt, 1989; Levelt & Wheeldon, 1994) and on the model of Van der Merwe (1997).

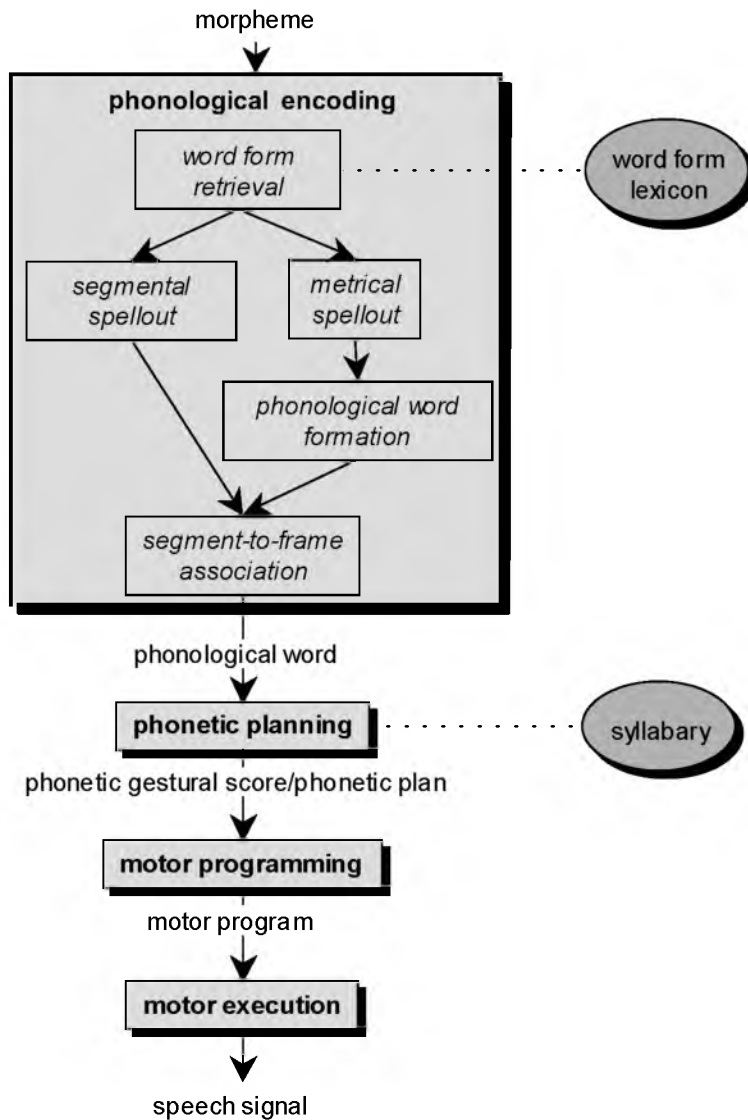


Figure 1: Model of speech production based on the models of Levelt and colleagues (latest model Levelt et al., 1999) and Van der Merwe (1997).

Phonological encoding

According to Levelt (1989), phonological encoding starts with retrieval of the word-form ('lexeme'). Phonological encoding comprises the spelling out of the word's metrical and segmental properties and inserting the segments in the metrical template, resulting in a phonological plan (Levelt et al., 1999). Deficits at this level may range from underspecified or incorrect lexemes to an inadequate or delayed phonological rule system that is different from both the target adult form and the age-appropriate developmental level form. In clinical linguistic descriptions an error pattern that remains the same over different events, for example the consistent use of non-developmental (atypical) rules, is often interpreted as reflecting these underlying deficits. In addition, one might find phonotactic errors and phoneme sequencing errors, such as sound substitutions (not distortions perceived as substitutions) and transpositions,

including metathesis. For example, 'bath' pronounced as 'path' results from an error at the level of phonological encoding when the wrong (first) phoneme is selected. However, this error might also concern a distortion (rather than a substitution) that emerges at a later stage in speech production (see Motor programming), namely when voicing in the first phoneme (which differentiates /b/ from /p/) is initiated too late and the /b/ is consequently perceived as /p/. Since phonological planning includes planning of suprasegmental features, prosodic disturbances may also result from this level (Ozanne, 1995; Van der Merwe, 1997).

Mental Syllabary

The mental syllabary, as proposed by Crompton (1982 in Levelt et al., 1999, p. 32) and adopted by Levelt (1989; Levelt & Wheeldon, 1994; 1999), is a repository of gestural programs of frequently used syllables that are retrieved during phonetic planning (which process will be discussed below). Making use of the syllabary has the advantage that the motor plan of a frequently uttered syllable needs not to be computed time and again, but is stored and can be retrieved on demand. Thus conceived, the syllabary can be interpreted as a mechanism for automaticity to ensure fast and effortless production (Varley & Whiteside, 2001; Ziegler, 2001). In addition, it is suggested that there is more coherence of spatial and temporal aspects of articulatory gestures within the syllables than between syllables (Browman & Goldstein, 1997). Syllables that are stored in a syllabary are likely to show more cohesion. A problem in accessing the mental syllabary or restoring a (precompiled) gestural program might thus lead to prolongation and less cohesion of the sounds within the syllable.

Phonetic planning

The next step is translating a phonological plan into a phonetic plan (McNeil et al., 1997; Ozanne, 1995; Velleman & Strand, 1994). To achieve this, the spatial and temporal goals of the articulatory movements for speech sound productions (the phonetic plan) are either retrieved from a sensorimotor memory and adapted to the surrounding phonemes, or precompiled gestural syllabic programs are obtained from the syllabary. A breakdown at this level could cause difficulty in recalling or restoring the correct motor plans of specific phonemes resulting in groping behavior on verbal tasks. Also, enhanced differences in performance on voluntary speech versus more automatic, standardized utterances could occur, because the former, being produced 'on the fly', requires more contextual adaptation than the latter, which are overlearned. Children with a speech disorder at this level might be able to utter words spontaneously but unable to imitate them, or to produce a sound but unable to do so in the appropriate context. This is due to the specific inability in adapting phonemes to the phonetic context arising from a disorder at this processing level (e.g. a child aged 3;6 years who produced 'car' as [da], but 'dog' as [pɒk]; Ozanne, 1995, p. 108). Thus, investigation of the articulatory cohesion within the syllable (intrasyllabic coarticulation) could provide information about a possible problem in phonetic planning (Van der Merwe, 1997).

Motor programming

Unlike Levelt and colleagues (Levelt et al., 1999), who suggested an articulatory network as the last stage of speech production in which the exact movement trajectories of the articulators are calculated, others (e.g., Ozanne, 1995; Van der Merwe, 1997) subdivided this last level into two stages: motor programming and motor execution (also see Figure 1). During motor programming the (more abstract) phonetic plans, that is the articulatory ‘gestures’, are translated into precise articulatory instructions, in a so-called ‘task dynamical system’ (see Browman & Goldstein, 1997; Fowler & Saltzman, 1993). This means that the gesture (defined during phonetic planning) only defines the task of the articulators in an abstract way and does not delineate the exact means to accomplish this task. For example, one of the tasks in producing the consonant /p/ is ‘lip closure’. The execution can consist of movements of the mandibular, the lower lip, both lips, or combinations of these articulators. The information required to reach the set goal is not specified until the motor programming stage.

The motor programming stage also allows compensation, for example, it permits speakers to still produce intelligible speech while clenching a bite-block between the teeth. A malfunction in motor programming affects the process of specifying muscle tone, rate, direction, and range of movements (Van der Merwe, 1997), resulting in problems such as sound distortions, voicing errors, resonance inconsistencies, or phonetic variability of production. Furthermore, a fine motor dyscoordination might result in slow diadochokinetic rates and the inability to maintain syllable structure, by producing perseverative responses (Ozanne, 1995). The two deficits of poor compensation (in e.g. a bite-block speech condition) and dyscoordination (slow diadochokinetic rates) could interact resulting in complex context-dependent speech patterns (see also Towne, 1994).

Motor execution

In the final stage of motor execution the motor program is transformed into automatic (reflex) motor adjustments, that is, it is implemented by the muscles involved in articulation. Problems at this stage might be due to an anatomical anomaly, such as cleft palate, or to neurological damage affecting muscle strength and coordination (dysarthric qualities).

POSSIBLE UNDERLYING DEFICITS IN DAS

How do possible underlying deficits of DAS fit into the model sketched above? Diverse explanations for the frequently noted unintelligible speech of children with DAS range from an impairment in storing and retrieving word forms, in producing the correct sequence of speech sounds in syllables and words (phonological encoding), in automating speech patterns such as syllables, to deficient phonetic planning and motor programming. Most authors agree that DAS is not caused by an oral-motor deficit like dysarthria, although a concomitant dysarthria is possible.

A core feature of DAS is *inconsistency*. Inconsistency of articulatory errors suggests a processing rather than a representational deficit; no consistent use of atypical phonological behavior has been reported for DAS that would indicate a common underlying phonological

representation problem. Also the fact that most children with DAS (from age 5 onward) do not demonstrate consistent problems in producing phonemic contrasts on request suggests that DAS can occur with a complete and intact *phonological repertoire*.

Furthermore, there is evidence that the origin of the speech symptoms in children with DAS is to be found in a stage following word-form retrieval, and is a processing rather than (phonological) representation problem (Thoonen, 1998). The evidence consists of studies that reported small differences in number of errors produced by children with DAS when imitating meaningful as compared to nonsense words, which was in contrast to normally speaking children who produced considerably more errors in nonsense words (Thoonen, Maassen, Gabreëls, & Schreuder, 1994). Following Ozanne (1995) we would argue that DAS arises from an impairment somewhere in the transition from word-form retrieval into the final articulo-motor output.

Phonetic transcriptions of utterances by children with DAS have revealed information about the type and amount of speech errors that these children produce in spontaneous speech as well as in repeated utterances. Although large *quantitative* differences between DAS and normally speaking children have been reported, namely higher substitution and omission rates, very few *qualitative* differences in error patterns have been found between children with DAS, children diagnosed with a phonological speech output disorder and normally speaking children (Forrest & Morrisette, 1999; Shriberg et al., 1997b; Thoonen et al., 1994). Thus, both the specificity of the phonological error patterns in children with DAS and the suggestion of the phonological encoding as *the* underlying deficit can be seriously questioned.

Table 1: Scheme of possible locations of the underlying deficit(s) in DAS.

Characteristics in DAS	Possible locations of the underlying deficit				
	phonological encoding	mental syllabary	phonetic planning	motor programming	motor execution
sequencing errors	+		+	+	
inconsistency in errors			+	+	+
small difference in numbers of errors					
imitating meaningful versus nonsense words		+	+	+	+
few qualitative differences in error patterns					
between DAS and NS			+	+	
in correct productions:					
prolongations of transitions			+	+	+
in correct productions:					
prolongations of steady states			+	+	+

Note. NS = normally speaking children

Given the fact that so few specific symptoms are found at the level of phonological encoding (also see Table 1) and their origin is presumed to lie after the word-form retrieval process, we hypothesize the following. Although children with DAS produce many phonemic errors, in phonemically *correct* productions in which it can be assumed that the phonological plan was

correct, we argue that children with DAS have problems in the transformation of the phonological plan into a phonetic plan and/or a motor program. This would then become manifest in qualitative differences in the speech productions of children with DAS as compared to the trouble-free productions in normally speaking children. These qualitative differences in phonemically correct utterances cannot be perceived auditorily, and therefore require a different methodology; acoustical measurements do allow these differences in articulation to be established. For example, prolongation of transitions, steady states, and intrasyllabic pauses, all characteristic of DAS, can easily be assessed acoustically, but are difficult to recognize perceptually; they “escape the ear”. Acoustic analysis of the speech may thus provide further indications with respect to the location of the underlying deficit in DAS (from phonological encoding onward) and may allow a distinction to be made between a deficient phonetic or syllable plan, a problem in the mental syllabary, or a deficit in motor programming.

COARTICULATION

In what way could acoustic measurements, then, contribute to the search for the underlying deficit in DAS? The articulatory realization of a segment is highly dependent on the phonetic environment, leading to coarticulation due to preplanning (i.e., anticipatory coarticulation) or carry-over (i.e., perseveratory coarticulation) effects. This means that before a phoneme is actually uttered, features of this phoneme (e.g. spectral quality and duration) can influence the preceding phonemes (Whalen, 1990). Problems in planning or in programming speech movements could influence the syllabic coherence and leave their traces in anticipatory coarticulation patterns. The influence of an upcoming vowel in phonemes preceding the vowel can be determined by measuring the formant frequencies throughout the utterance. For example, the high second formant frequency of an upcoming /i/ versus the low second formant frequency of an /o/ can be found earlier in an utterance (for instance in /zə#sxit/ [‘she shoots’] versus /zə#sxot/ [‘she shot’] as described in one of the experiments of this thesis), reflecting anticipatory coarticulation. Thus, investigating the coarticulation pattern and contextual interdependency in utterances might provide us valuable information about possible problems in planning or programming speech.

Before discussing the experiments we conducted in order to find answers as to the underlying deficit in DAS, I will first discuss the results of studies concerning coarticulation in normal development and in adults with acquired apraxia of speech.

Coarticulation in normal speech development

What do we know about coarticulation effects in normal speech development? Developmental studies show divergent results on the programming of syllables and segments by adults and children. In a series of coarticulation experiments with bisyllabic words of the type /əCV/ (C=/s, ʃ, t, k, d/, and V=/æ, i, u/), Nitttrouer and colleagues (Nitttrouer, Studdert-Kennedy, & McGowan, 1989; Nitttrouer, 1993; Nitttrouer, Studdert-Kennedy, & Neely, 1996) tried to shed more light on this issue. They showed that in normal speech development coarticulation between

syllables (intersyllabic coarticulation: effect of V on [ə]) diminishes at an earlier age than coarticulation within syllables (intrasyllabic coarticulation: effect of V on C). Accordingly, children's speech records show more intersyllabic coarticulatory cohesion, and in speech development, the reduction of intrasyllabic coarticulation lags behind the reduction of intersyllabic coarticulation. These results could be interpreted as evidence that children initiate the following vowel gestures earlier in the utterance than adults do, which supports the view that the phonetic segment is the endpoint rather than the starting point of development. Like Nitttrouer et al. (1989; 1993; 1996), Siren and Wilcox (1995) found more coarticulation in children's speech than in adult speech. Additionally, they showed that the primary coarticulation effect is intrasyllabic, and that the effect of the vowel on the preceding fricative is larger in nonsense utterances than in meaningful utterances.

Conversely, some researchers propagate an alternative view maintaining that during development the skill of syllable cohesion is learned after the children have mastered the articulation of the individual segments (Kent & Rosenbek, 1983; Sereno, Baum, Marean, & Lieberman, 1987; Sereno & Lieberman, 1987). From this perspective, the speech of children should reveal less evidence of coarticulation within and between syllables than the acoustic records of adults. Sereno and Lieberman (1987) showed that lingual anticipatory coarticulation on [k] preceding [a] or [i] was more apparent in adults' than in children's utterances, both acoustically and perceptually. This finding was supported by studies of Sereno, Baum, Marean, and Lieberman (1987), in which adult speech utterances showed stronger acoustic and perceptual cues of anticipatory labial coarticulation than children's utterances. The above shows that the results of coarticulation research on normal speech development are rather contradictory.

Coarticulation in acquired apraxia of speech

Like the contradictory results that have been reported in studies on normal development of coarticulation, studies concerning coarticulation in adult apraxic patients describe similar divergent results (Dogil, Mayer, & Vollmer, 1996; Southwood, Dagenais, Garcia, & Sutphin, 1996; Ziegler & Von Cramon, 1985; 1986). In a study of labial coarticulation (utterances as /gəʔVtə/, in which V=/i,y,u,a/) Ziegler and Von Cramon (1985; 1986) found a lack of coarticulatory cohesion in a patient with apraxia of speech, which seems to reveal a problem in appropriately phasing speech gestures. Dogil, Mayer, and Vollmer (1996) found a similar absence of coarticulatory cohesion. They explained the lack of coarticulatory cohesion in terms of phonological overspecification. On the other hand, the data obtained by Southwood et al. (1996) showed preservation of lingual coarticulation (in utterances like 'say sheet again' versus 'say shoot again') in adults with apraxia of speech at slow and habitual speaking rates but delayed or distorted coarticulation at fast rates. Contrary, the onset of labial coarticulation (in utterances like 'say beat again' versus 'say boot again') was delayed in the apraxic speaker at all speaking rates. Katz and colleagues (1987; 1988) did not find any differences between normally speaking adults and adults with apraxic speech characteristics with respect to anticipatory coarticulation. In the studies of the present thesis, coarticulation plays a central role.

OBJECTIVES AND OUTLINE OF THE STUDY

Thus, the aim of the studies reported in the present thesis was to gain more insight into the underlying deficit(s) in DAS. In order to investigate whether children with DAS have problems in the planning and/or programming of speech, we conducted a series of experiments. This thesis comprises a description of unique, experimental studies in which the research strategy was as follows. Firstly, each level of the speech production process that might be impaired in DAS was studied independently in separate experiments. Secondly, we restricted ourselves to the comparison of the speech production in children with DAS with normally speaking children (note that this did not comprise differential diagnoses with other speech disorders). Thirdly, the utterances of the normally speaking children were compared to those of adult women in order to address both the issue of development in coarticulation and possible deviant coarticulation. Finally, acoustic measurements were conducted on the correct utterances of children with DAS and those of normally speaking children.

In order to select a group of children that were clear cases of DAS we used stringent selection criteria. Children with additional problems (like language comprehension problems, dysarthria, hearing problems, and below average intelligence) that might obscure any conclusions were excluded. A detailed description of the selection procedure is given in the following chapters.

Besides the speech data, we also collected data concerning cognitive, including executive, functions of children with DAS. Problems in programming sequences of movements might not be restricted to speech production and might also appear in other movements, indicating a more generalized problem. Alternatively, one could speculate that the capacity to generate sequences from memory governs the ability to sequence movements. By collecting data of cognitive functions we addressed the issue of a possible generalized deficient mechanism as the underlying cause of DAS. Since cognitive functions were tested twice within a timeframe of 1;3 years the developmental aspect was also considered. By doing so, the results obtained also contribute to the theoretical discussion of DAS as a delay or deviance in development.

DAS, a problem in phonetic planning?

As a first hypothesis for the underlying deficit in DAS we studied the possibility of deviant phonetic planning. For this purpose we first conducted a study into the articulatory cohesion within and between syllables, by which the involvement of planning (and/or programming) of speech movements was evaluated in various contexts. Chapter 2 describes the experiment we conducted in order to establish coarticulation patterns in children with DAS in a relatively easy speech-production task. CV syllables were produced in which the coarticulation effects of a varying vowel (V) were measured earlier in the utterance, that is in the preceding consonant (C) and in the preceding syllable in the schwa.

Based on the findings that cohesion of articulatory movements within syllables is stronger than between syllables (Levelt et al., 1999; Whalen, 1990) we argued that articulation is organized on the basis of a stored repository of syllabic gesture scores. As a consequence, the syllable structure of a particular utterance is determinative of the amount of segmental coarticulation.

Therefore, we manipulated the syllable structure in an otherwise unchanging sequence of sounds. An example in English is ‘I scream’ versus ‘ice cream’ in which the phonemic sequence is identical in both utterances. If the syllable structure is indeed relevant to the amount of coarticulation, then, in this example, the anticipatory coarticulation of the vowel on the preceding /s/ is expected to be stronger in the first phrase (within syllabic coarticulation) than in the second phrase (between syllabic coarticulation). In the experiment described in Chapter 3 similar manipulations were applied. On the basis of the assumption that DAS is a problem of phonetic planning, we expected to find different effects of the syllable structure manipulation in children with DAS as compared to normally speaking children.

DAS, a problem in accessing or storing the mental syllabary?

In addition to syllable structure, the experiment described in Chapter 3 also looked at the contrast between high-frequent syllables and syllables with an extremely low (zero) frequency. The assumption is that a necessary (but not sufficient) condition for storage of syllables in the syllabary is their frequent use in speech. Syllables that are used frequently in the ambient language are therefore more likely to be stored in the syllabary than low-frequent syllables. Even if a child is in the process of building a syllabary, we may expect an effect of syllable frequency. If a child is unable to store syllables or to access the syllabary, no difference in coarticulation is expected between high and low-frequent syllables.

DAS, deviant motor programming or motor execution?

In order to explore whether in DAS a deviance might occur during the final stages of speech production we compared the ability of normally speaking children and children with DAS to compensate their articulatory movements for perturbations (Chapter 4). Such a compensation is possible from the level of motor programming onward (Van der Merwe, 1997). In the experiment described in Chapter 4, children were asked to produce utterances in a condition in which their mandible was fixed by a bite-block clenched between their teeth in such a way that vertical articulatory movements were virtually made impossible. These utterances were compared to their speech productions in a condition without bite-block. It was our premise that problems in compensating articulatory movements in such a bite-block condition might reveal an underlying disruption in motor programming. We expected the children with DAS to show more problems in adapting to the bite-block condition than the normally speaking children.

DAS, a problem in using stress?

An alternative hypothesis was put forward by Shriberg et al. (1997a), who reported that children with DAS especially have problems in stress resulting in misplaced or equal stress throughout the utterance (so-called ‘scanning speech’ in which problems arise in stressing and unstressing appropriate syllables in a word and words in a phrase). These authors interpreted the deviating durational patterns in the utterances of children with DAS as resulting from problems in rhythm and prosody, which, rather than reflecting differences in movement durations (or ‘prearticulatory

sequencing’), correspond to deficiencies in durational control. These findings are in line with Manuel (1999) who concluded: ‘... prosody clearly has to do with timing, so it seems likely that prosody and temporal coordination of articulatory gestures are strongly linked’ (p.196). In Chapter 5 we studied the durational patterns in the speech of children with DAS in order to evaluate durational control. By investigating durational control we also expected to answer the question whether slow speaking rate, as commonly found in children with DAS, reflects the underlying disorder or can be considered a compensatory strategy. For this, segment durations and contextual interdependency were investigated in a normal speech condition and in a compensatory, that is bite-block, speech condition.

DAS, a problem of more than speech alone?

In the introduction I mentioned the fact that children with DAS also often demonstrate ‘soft’ neurological signs such as clumsiness and motor coordination problems (Guyette & Diedrich, 1981; Ozanne, 1995; Pollock & Hall, 1991; Robin, 1992). Sequencing problems might not be restricted to speech production but might appear in other movements as well and may even manifest themselves in sequential memory tasks. In Chapter 6 we investigated cognitive functions, including executive functions, in children with DAS in order to evaluate whether other functions besides speech production are impaired in DAS. Besides studying sequential abilities, both motoric and auditory, and sequential memory, we also explored sensory functions, sensory integration, motor repetition and spatial memory. Furthermore, children with DAS were compared with normally speaking children on two measurements (1;3 years between both measurements), in order to evaluate whether possible weaker scores of children with DAS are due to either a delay or a deviance in development.

Finally, in the general discussion in Chapter 7 the results of the experiments are summarized and discussed in the light of the study’s objectives.

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CHAPTER TWO

Coarticulation patterns in DAS

Nijland, L., Maassen, B., Van der Meulen, Sj., Gabreëls, F., Kraaimaat, F.W., & Schreuder, R. (2002). Coarticulation patterns in children with Developmental apraxia of speech. *Clinical Linguistics and Phonetics*, 16 (6), 461-483.

ABSTRACT¹

The aim of this study was to enhance our insight into the underlying deficit in developmental apraxia of speech (DAS). In particular, the involvement of planning and/or programming of speech movements in context was tested by analysing coarticulatory cohesion. For this purpose, second formant frequency measurements were conducted in repetitions of nonsense utterances ([əCV] C=/s,ʌ,b,d/; V=/i,a,u/), and compared across nine children with DAS, six normally speaking (NS) children, and six adult women. The results showed both intra- and intersyllabic anticipatory coarticulation in NS children and adult women, in which the intersyllabic coarticulation was stronger in NS children than in adult women. The children with DAS showed more variability as compared to NS children, made, on average, less distinction between the vowels, and showed individually idiosyncratic coarticulation patterns. These results are discussed in the light of a delay as well as a deviance of speech development in children with DAS.

INTRODUCTION

Developmental apraxia of speech (DAS) is an impairment that leads to a serious communicative disability. There is debate in the speech literature regarding specific characteristics of DAS. Early research was mainly focussed on the phonemic descriptions of the erroneous production of children with DAS. Since the end of the eighties more phonetic research has been conducted to study underlying processes (e.g., Hall, Jordan, & Robin, 1993). In the present study we attempt to gain more insight in the underlying deficit of DAS by measuring the phonetic coarticulation effects in correctly produced utterances.

Characteristics of DAS

There has been much discussion about the issue whether DAS can be seen as a diagnostic entity. One of the main issues is the overlap between DAS and other speech disorders. McCabe, Rosenthal and McLeod (1998) found that many characteristics regarded as diagnostic for DAS also occur in the general speech-impaired population. The debate continues because attempts to find a diagnostic marker specific for DAS, up to now have not been successful (Shriberg, Aram, & Kwiatkowski, 1997). Despite the overlap with other speech disorders and diagnostic difficulties, there is agreement about some of the more central or core characteristics of DAS. The speech is often unintelligible due to a large number of phonemic speech errors (substitutions and omissions) and articulatory abnormalities. These speech errors are inconsistent and the number increases with increasing complexity and length of the utterance. Spontaneous speech production is generally more disrupted than imitative speech. The most salient speech characteristics of children with DAS are: largely unintelligible speech, sequencing errors, abnormal prosody, high consonant error rates, many context-related substitutions, groping, and inconsistency of errors (Hall et al., 1993; Thoonen, Maassen, Wit, Gabreëls, & Schreuder, 1996). Non-speech actions, like coughing, chewing and swallowing, do not necessarily cause difficulties.

¹ The journal in which this chapter is published uses British-English spelling therefore this chapter has British-English spelling rather than American-English.

The number of studies concerning DAS is much smaller than the amount of studies about apraxia of speech in adults (AOS). Although both disorders arise from different origins, and there are differences between the speech characteristics of AOS and DAS, on the methodological level much can be learned of the study of the one compared to the other. Most research on AOS in adults and DAS has been conducted on the basis of perceptual evaluations, i.e. phonemic descriptions of segmental speech errors, which can be extended with analyses on the feature level (Forrest & Morrisette, 1999; Thoonen, Maassen, Gabreëls, & Schreuder, 1994). However, perceptual analyses have their methodological limitations. The relatively subtle phonetic differences (for example tongue displacement) remain inconspicuous in perceptual evaluations. Instrumental analyses provide quantitative, objective data on a wide range of different speech parameters that go beyond the scope of an auditory-based judgement (Hardcastle & Edwards, 1992), as was shown in studies on the acoustic characteristics of the speech of subjects with AOS. Results showed that subjects with AOS differed from normally speaking subjects in voice onset time distribution by compression of the two categories of voiced versus voiceless stops (Freeman, Sands, & Harris, 1978; Hoit-Dalgaard, Murry, & Kopp, 1983; Itoh et al., 1982), in longer word and vowel durations with prolongation of transitions, steady states, and intersyllabic pauses (Caligiuri & Till, 1983; Collins, Rosenbek, & Wertz, 1983; Kent & Rosenbek, 1983), and in reduced intensity variations (Kent & Rosenbek, 1983). In contrast to AOS, only few studies report acoustic characteristics of *developmental* apraxia of speech. As a consequence, very little is known of the phonetic details of the speech of children with DAS.

Coarticulation

What is the underlying deficit in DAS? Several speech characteristics suggest that the underlying deficit should be found in an impairment of planning and/or programming of speech movements in context. From a motor perspective the articulatory unit is not a single phonemic segment, which means that the phonemic motor patterns are not invariant. Rather, successive articulatory gestures are highly dependent on the phonemic context and extend across phonemes, which results in articulatory overlap or coarticulation (Browman & Goldstein, 1992). If a segment influences a following segment this is called perseveratory coarticulation and an upcoming segment influencing a preceding segment is called anticipatory coarticulation. In this motor view, problems in planning and programming of speech movements leave their traces in the coarticulatory cohesion of the utterances. Hertrich and Ackermann (1995), for example, found that in normal speech slowed speech rate resulted in a decrease of perseveratory coarticulation and, against their expectations, unaltered or even increased anticipatory coarticulation. From this, they suggested that different mechanisms underlie anticipatory and perseveratory coarticulation. Thus, coarticulation measurements yield valuable data regarding speech motor processes. Therefore, in the present study we investigated the nature and amount of anticipatory coarticulation in utterances of children with DAS in order to determine a problem in planning and/or programming of speech.

Although hardly any research was done on coarticulation patterns in children with DAS (e.g. Sussman, Marquardt, & Doyle, 2000), studies on coarticulation in AOS in adults are mentioned frequently. These studies have reported divergent results. Whereas some researchers found a lack

of coarticulatory cohesion in apraxic patients (Dogil, Mayer, & Vollmer, 1996; Whiteside & Varley, 1998; Ziegler & Von Cramon, 1985; 1986), others did not (Katz & Baum, 1987; Katz, 1988). The data of Southwood, Dagenais, Garcia and Sutphin (1996) showed that speaking rate and articulators involved might be one of the sources of the different coarticulation patterns in apraxic speech (delayed coarticulation at fast rates and labial coarticulation). Furthermore, divergent problems underlying AOS were suggested, varying from inappropriately phasing speech gestures (Ziegler & Von Cramon, 1985; 1986), and lack of automation (Whiteside & Varley, 1998), to phonological overspecification (Dogil et al., 1996).

In order to overcome interpretation problems due to differences in speech tasks and material, the design of the present study included both lingual and labial consonants and besides the research group, children with DAS, also two control groups were included (normally speaking children and adult women). Considering the divergent results on AOS, we might expect either *weaker* or *stronger* coarticulation in children with DAS as compared to normally speaking children, in the present study. On the one hand *weaker* coarticulation could be predicted in DAS based on the frequently reported slow and protracted speech, due to a protracted segment-to-segment motor planning or programming. This would result in a lack of coarticulatory cohesion. On the other hand, *stronger* coarticulation effects (more influence of upcoming vowel on preceding sounds) could be found in the speech of children with DAS as a result of a more global planning of the utterances, under the hypothesis that motor control is not fully developed. If such were the case, then DAS speech would resemble the speech of younger, normally speaking children, which is characterised by *stronger* coarticulation (Nittrouer, Studdert-Kennedy, & McGowan, 1989; Nittrouer, 1993; Nittrouer, Studdert-Kennedy, & Neely, 1996).² Furthermore, in the present study, it is considered whether these coarticulation effects are restricted to the sounds within the syllable, called 'intrasyllabic coarticulation', or extend across the syllabic boundary, 'intersyllabic coarticulation' (Boers, Maassen, & Van der Meulen, 1998). The above-mentioned pathological and developmental studies on coarticulation will be discussed in more detail in the Discussion, together with the findings of the present study.

The methods adopted in the present study were derived from the studies of Nittrouer and colleagues (Nittrouer et al., 1989; Nittrouer, 1993; Nittrouer et al., 1996). In [əCV]-utterances we measured the influence of the vowel V on the preceding consonant C (intrasyllabic coarticulation), and on the schwa [ə] (intersyllabic coarticulation). Three different vowels (/i, a, u/) were chosen that are the extreme vowels in the Dutch vowel space. These vowels are distinctive in second formant (F2) frequency value. The influence of the vowel on preceding segments (anticipatory coarticulation) was determined by measuring the F2 values throughout the utterance. The coarticulation patterns (the F2 patterns) of children with DAS were compared to those of normally developing children.

² Note that, a similar reasoning can be made for the coarticulation patterns in other speech disorders. Since no other speech disorders are being considered in this study, we do not intend to draw strong conclusions as to the specificity of the findings for the speech disorder DAS, as compared to, for instance, phonological disorders.

Thus, the question for this study was, does the speech of children with DAS show *weaker* or *stronger* coarticulation as compared to normally speaking children? And, if so, are these effects restricted to the syllable (intrasyllabic) or do they cross the syllable boundary (intersyllabic)?

METHOD

Participants

The acoustic analyses were rather time consuming, therefore only nine children with DAS and six NS children were analysed. We decided to analyse a greater number of children with DAS than NS children, since their individual patterns were expected to be more diverse. The participants were randomly taken from 19 children with DAS (14 boys and 5 girls between the age of 4;11 and 6;10) and 19 NS children (matched for sex, age, and dialect region). All children were native speakers of Dutch.

The children with DAS were selected from special schools for children with speech and language disorders. Speech-language pathologists of these schools referred 70 children diagnosed as having dyspraxic speech problems to us. Based on samples of spontaneous speech, repetitive imitations of words and brief phrases, and a diadochokinetic task (collected by the school speech-language pathologist) the following selection criteria were adopted: exhibiting many phonemic errors despite a complete phoneme repertoire, high frequency of consonant substitutions (and omissions in clusters), sequencing difficulties of phonemes and syllables, inconsistent error patterns, and inability to produce complex phonemic sequences (Hall et al., 1993; Thoonen et al., 1996). To the selection of 29 children from the larger group of 70 children the following tests were administered: assessment of hearing level, a language comprehension test (the Dutch version of the Reynell Developmental Language Scales; Bomers & Mugge, 1989; Reynell & Huntley, 1985), and speech tasks developed by Thoonen et al. (1996). Furthermore, it was established that the children did not exhibit organic disorders in the orofacial area, gross motor disturbances, dysarthria, or below-normal intelligence. Based on the test results, another 10 children were excluded because of hearing problems and poor language comprehension (test score 1 standard deviation or more below average). The remaining 19 children can be considered as ‘clear’ cases of DAS.³

In previous studies we found that the speech of children with DAS is particularly characterised by (1) a high consonant substitution rate, which is not only high in nonsense utterances but (2) also in meaningful speech, and (3) difficulty with the Maximum Repetition Rate task on tri-syllabic sequences (‘pataka-pataka..’) as compared to mono-syllabic sequences (‘papa..’, ‘tata..’, ‘kaka..’) (Maassen, Thoonen, & Boers, 1997; Thoonen et al., 1994). Therefore, these parameters were extracted from the speech material collected from the administered speech tasks. (For the exact procedure of administering this assessment the reader is referred to Thoonen et al., 1996). The results are presented in table 1. The percentages consonant substitution of singleton

³ Obviously, all children who were referred by the speech-language pathologists showed apraxic characteristics, however, only those who satisfied all criteria were included in the present study. Including only those children who can be considered as ‘clear’ cases of DAS enables us to study the nature of DAS.

consonants on syllable initial position ('Subst'), and from these the percentage of substitutions with respect to place-of-articulation ('Substpl'), were derived from an imitation task of meaningful (first two columns of table 1) and nonsense utterances (third and fourth column of table 1). Maximum Repetition Rates (MRR), expressed in mean number of syllables per second, are given for the monosyllabic (the average of the three target monosyllable utterances, 'papa..', 'tata..', or 'kaka..') and trisyllabic ('pataka..') sequences in column five and six. In case the child was not able to produce five successive trisyllabic sequences correctly, even after several attempts, this is indicated with 'Unable'. The data in the last column of table 1 are explained below. The percentages consonant substitution and the MRR scores clearly distinguish the two groups: the children with DAS showed higher percentages consonant substitution – remarkable are the percentages substitution in meaningful utterances and the substitution of place – and smaller MRR scores as compared to the NS children.

Table 1. Individual scores on the selection tasks (percentage consonant substitution in meaningful and nonsense word-imitation task and mean number of syllables per second in Maximum Repetition Rate task), and on the experimental task (percentage correctly produced utterances) in children with DAS and normally speaking children (NS).

DAS	Age	Meaningful		Nonsense		MRR		% correct
		Subst	Substpl	Subst	Substpl	Mono-syll	Tri-syll	
#1	5;0	26%	76%	47%	68%	3.53	1.93	82
#2	5;1	24%	56%	64%	79%	4.12	3.38	89
#13	5;6	22%	71%	38%	68%	3.64	2.21	64
#14	5;7	15%	60%	15%	30%	4.63	3.68	96
#17	5;10	17%	64%	67%	70%	4.63	Unable	89
#20	5;11	30%	60%	48%	72%	3.31	Unable	61
#21	5;11	31%	85%	-	-	3.20	Unable	96
#28	6;10	40%	60%	39%	96%	4.23	3.61	57
#29	6;10	14%	56%	15%	60%	3.98	Unable	86
NS	Age	Subst	Substpl	Subst	Substpl	Mono-syll	Tri-syll	% correct
#54	4;9	7%	0%	12%	38%	4.29	3.52	90
#36	5;0	0%	0%	17%	64%	4.85	3.68	92
#42	5;3	3%	0%	6%	25%	4.74	4.05	93
#53	5;6	8%	0%	8%	20%	4.49	5.03	100
#58	5;6	5%	33%	9%	33%	4.44	5.08	96
#49	5;11	1%	0%	13%	0%	4.97	3.06	100

Note. Subst: percentage substitution of singleton consonants on syllable initial position. Substpl: percentage substitution of place relative to the number of substitutions (Subst). Mono-syll: mean number of syllables per second of monosyllable utterances (/pa/, /ta/, /ka/). Tri-syll: MRR in a tri-syllable utterance /pataka/.

Apart from the children, also six adult women were analysed. They functioned as a reference of the end stage of normal development, were all between 20 and 30 years of age, and had no history of speech pathology, orthodontics, and hearing problems. We opted for women because,

rather than male voices and vocal tracts, female voices and their vocal tracts are more similar to those of children.

Speech material

The speech samples consisted of disyllabic nonsense utterances of the type [əCV], in which V was one of the extreme vowels /a, i, u/ of the Dutch vowel space, and C was either a fricative (the alveolar /s/ or the velar /x/) or a stop (/b/ or /d/). These utterances are comparable to the speech material of Nittrouer et al. (1989; 1993; 1996). However, in contrast to the alveolar-palatal distinction in the fricatives /s/ - /ʃ/ used by Nittrouer et al. (1989; 1993; 1996), the alveolar-velar distinction /s/ - /x/ was used in this study. The reason for this choice was that the articulatory positions are further apart, and the fricative /x/ is very common in Dutch, in contrast to /ʃ/. We decided to use voiced stops (/b, d/).⁴ The reason for this is that formant transitions can be determined better in voiced stops as compared to voiceless stops (Blumstein & Stevens, 1979). To compare the results of the stops with the fricatives on place of articulation, it would have been better to use the voiced stops /d/ and /g/. However, the /g/ does not occur in Dutch (only as a result of assimilation, or in loan words).

All items were spoken in the same carrier phrase, 'he de ... weer' [hed ... wɪ:r] ('hey the ... again'), and repeated six times, so in the end each child produced 72 utterances (3 vowels * 4 consonants * 6 repetitions). Sometimes children (especially the children with DAS) were not able to correctly produce an utterance. In that case a second attempt was made immediately after the first. A unidirectional dynamic microphone mounted on a headset (Shure SM10A) and a tape-recorder (Kenwood KX54) were used to record the speech samples. The headset kept the microphone at a constant distance of 5 centimeters in front of the right corner of the subject's mouth.

Acoustic analyses

Selection and digitizing

The speech samples were digitized with a sample frequency of 25 kHz and the relevant sections (i.e. schwa-C-V segments) were spliced out, using the Kay Elemetrics Computerized Speech Lab (CSL) analysis system, Model 4300B. During the sampling procedure, utterances of which the [əCV]-part was phonemically incorrect were skipped. This led to a reduction of the number of repetitions, never to a total absence of an utterance type. Although the reliability of this selection procedure was not formally tested, it was subject to consensus discussion during the determination of the reliability of the measurement procedure (see below). In the last column of table 1 the percentages of correct utterances per child are given, which show that children with DAS produced more phonemically erroneous utterances than NS children. For 7 out of 9 children with DAS the percentages correct were below 90%; all NS children produced between 90% and 100% correct.

⁴ The English and Dutch voiced stops /b/ and /d/ differ in voice-onset-time: In the Dutch voiced stops the voicing starts before the burst, in the English voiced stops the voicing and burst occur simultaneously.

As a first step in the acoustic analyses, information of the oscillogram, FFT-spectrogram, and energy window was used to determine the onset and offset of each segment (schwa, consonant, and vowel⁵), and markers were set at the segments' onsets and offsets and at the plosive burst, using indications given by Nittrouer (1993). Inter-observer agreement concerning the placing of these markers was tested in a subset of the data. For this purpose, three observers independently placed markers in the 72 utterances of two children, namely one child with DAS and one NS child. High correlation coefficients were found (from 0.78 to 0.99 over all markers), which indicates a significant reliability between the observers. The mean difference in marker position over all markers was 12.2 ms (SE=2.8 ms), which was larger in the utterances of the child with DAS (mean=17.2 ms; SE=5.6 ms) than in the utterances of the NS child (mean=7.6 ms; SE=0.9 ms). The markers at the onset and offset of the segments were used to determine F2 values at particular locations throughout the utterance.

Formant extraction

The second formant (F2) trajectory was used to determine the differences between utterance types. In the voiced sections of the signal (i.e. schwa and vowel) the CSL-program automatically positioned impulse markers just before a major amplitude peak and at a positive zero crossing (equals the closing of the glottis during voicing). However, sometimes this automatic placing was not accurate. Whenever necessary, the wrongly placed impulse markers were corrected interactively. After this, the formant values (with corresponding bandwidths) were obtained using pitch-synchronous Linear Predictive Coding (LPC) analyses (triangular analysis window; 20 components autocorrelation with pre-emphasis of 0.950). F2 values were extracted at five locations in the schwa and vowel: at schwa-midpoint (1) and schwa-offset (2), at the vowel transition onset (3) and at transition end (4), and at vowel midpoint (5). A separate LPC-analysis was performed on a Hamming window of 20 ms in the consonant (6) in accordance with Nittrouer (1993); in the fricatives the measurement window was centred at 20 ms before offset; in the plosives the start of window was at the plosive burst.

Measuring the first and second formant frequencies is quite hard in children's and female voices due to the high fundamental frequencies (Bennett, 1981). Therefore, a post-processing procedure was followed to reject invalid formants. For each of the four relevant vowels (schwa, /i/, /a/, /u/) lower and upper limits for the first (F1) and second formant (F2) were determined by inspection of the LPC-based formant values plotted as dots over the FFT-spectrograms. Only formant values that fell within the black regions in the spectrogram were accepted as valid. The mean midpoints and widths of the ranges for each vowel and group of speakers are presented in table 2. These ranges are quite large due to inter-speaker differences. Because of these large inter-speaker differences, upper and lower limits were determined for each individual speaker separately. These upper and lower limits were used in the next analysis step. To determine whether the frequency values, obtained with the LPC-analyses, fell within the range defined in the preceding step and thus could be considered the first and second formant, a computer-

⁵ Although in [əCV] both /ə/ and V are vowels, here, we will address the first vowel as 'schwa' and the second vowel as 'vowel'.

program was developed for post-processing. The post-processing procedure was conducted per analysis-frame, i.e. per impulse marker.

Table 2. Mean midpoints and width of ranges of F1 and F2 (in Hz) in the schwa and vowels of each group.

Utterance	Children with DAS		NS children		Adult women	
	Schwa	Vowel	Schwa	Vowel	Schwa	Vowel
[əCa] - F1	659; +/-351	945; +/-543	570; +/-253	905; +/-543	431; +/-155	730; +/-370
F2	2396; +/-853	2049; +/-618	2201; +/-698	2005; +/-597	1988; +/-378	1761; +/-423
[əCi] - F1	648; +/-339	567; +/-261	538; +/-238	532; +/-232	402; +/-133	381; +/-131
F2	2463; +/-830	2845; +/-712	2206; +/-441	2695; +/-700	2186; +/-418	2569; +/-473
[əCu] - F1	618; +/-283	573; +/-268	573; +/-273	502; +/-202	400; +/-136	470; +/-205
F2	2283; +/-945	1652; +/-894	2084; +/-787	1483; +/-880	1980; +/-503	1118; +/-600

Note 1. Ranges for schwa were determined separately for each utterance type, resulting in minor differences depending on the upcoming vowel. Note 2. Ranges displayed here are rather broad, which reflects the differences between speakers within and between groups. Precautions were taken to find the 'correct' formant within the ranges (see text).

As a first step in the post-processing procedure the frequency values were sorted from low to high, and all formants with a bandwidth higher than the predetermined maximum bandwidth, which was fixed at 600 Hz, were eliminated. In the next step it was decided whether the remaining formant values fell within the F1- or the F2-ranges. If a value was found within the F1-range, this value was accepted as valid F1 value. In case more than one value was found, the higher one was taken, in order to be sure that F0 was not mistaken for F1. At the same time, the lower limit of the F2-range was shifted downward (in the current analysis frame) to the found F1. In the same way, a formant frequency within the F2-range was accepted as valid F2 value. If more than one formant fell within the F2-range, the lower one was taken, in order to avoid mistaking F3 for F2.

The small variance of second formant values that was found in the repeated utterances of the adult women and of the NS children (which will be discussed later in table 3), made us conclude that the procedure was quite reliable. Measurement of the first formant frequencies resulted in many missing values, since the distinction between the fundamental frequency and the first formant could not always be made appropriately. Also in the second formant frequency measurement using this procedure suffered from missing values. However, the percentage of accepted F2 values was similar across the three groups, viz. the smallest was found in the consonant (DAS 73%; NS 80%; AW 66%) and the largest at vowel midpoint (DAS 99%; NS 99%; AW 94%).

Large inter-speaker variance in mean formant values was expected, partly due to anatomical differences between speakers. To correct for this variability, as a means of speaker normalisation, formant *ratios* were calculated for each child separately. For each consonantal context, the formant values (of 6 repetitions) of utterances with /i/ were averaged and divided by the

averaged formant values of utterances with /u/ (i/u-ratio).⁶ Figure 1 gives a stylised example of a formant pattern and the calculated ratio-pattern. These ratios reflect F2-distinctions between the utterances. Typically, large ratios are found at vowel midpoint, where the distinction between the utterances is at maximum. Smaller ratios close to unity, such as typically found in the schwa, reflect that F2 values are similar despite differences in the upcoming vowel. Thus, the higher the ratio is above 1 the more distinction there is between the utterances at that particular location.

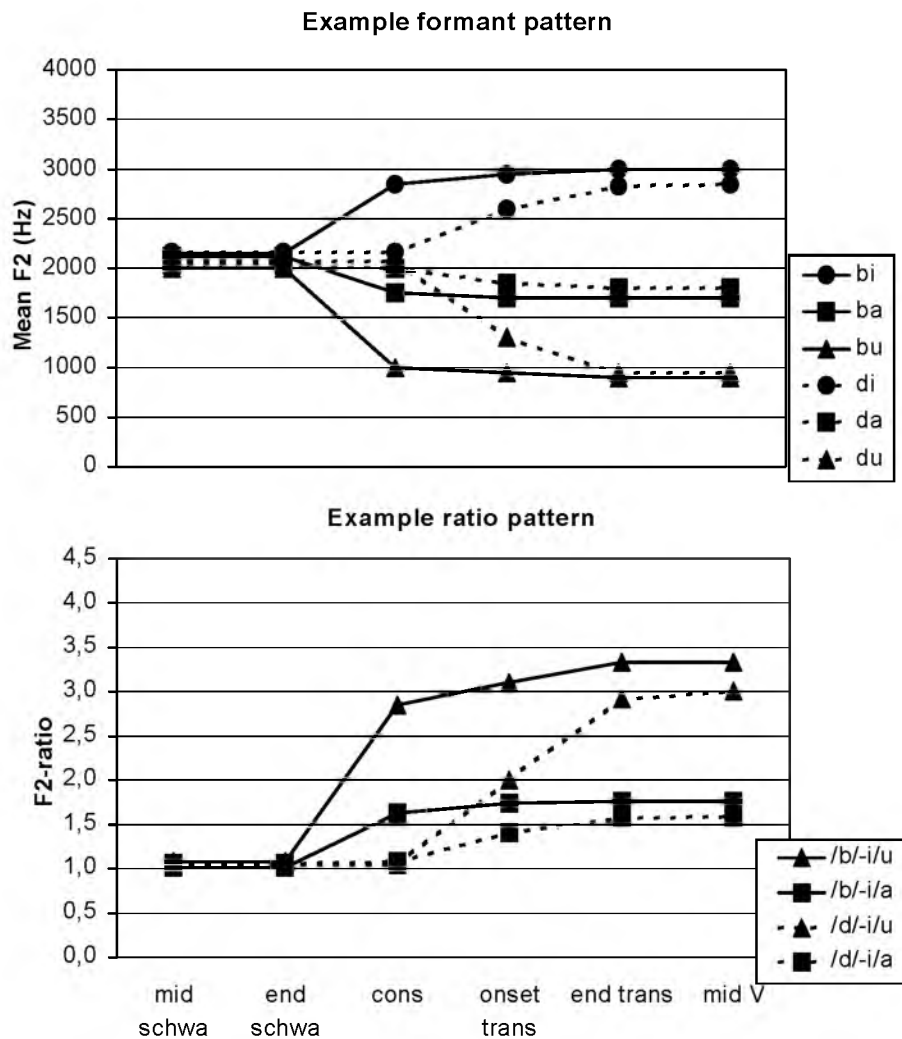


Figure 1. A fictitious example of the pattern of F2 values throughout the utterance (upper figure) and the calculated ratios.

⁶ Also i/a-ratios were calculated. Overall, the value of the i/a-ratios was lower than of the i/u-ratios, as expected, but their patterns were similar. The i/a-ratios are not presented in the figures.

Statistical design

In order to evaluate the statistical significance of formant value differences, analyses of variances were conducted first. However, parametric testing was not justified, because for most contrasts Levene's test for homogeneity of variance turned out to be significant (Winer, Brown, & Michels, 1991). Therefore, nonparametric tests were conducted. Another way to solve this problem of heterogeneity is to aggregate the repeated utterances, resulting in average formant values of each utterance type per child. Subsequently, F2 ratios were calculated and an analysis of variance was performed on the obtained data. Eta-square (η^2) was calculated to determine the effect size of the analyses of variance. In order to test whether the F2 ratios were significantly higher than 1, a one-sample *t*-test was performed.

RESULTS

In this section, before presenting the results of formant frequency measurements, the variability will be given. Above, in table 1, it was discussed that the scores of MRR and the percentages correct utterances of the children with DAS were more variable than those of NS children. Therefore, the variability within and between speakers per group is expected to provide valuable data. In table 3 the standard deviations of the formant frequencies are given, subdivided in variability attributable to the between subject factor 'Speaker' and the within subject factors 'Type' (utterance type) and 'Error' (the variability within speakers that is not attributable to the vowel or the consonant, but to the repetition of the same utterance). *F*-ratios were calculated to determine the significance of the difference in variance (viz. the square of the standard deviation presented in table 3) between the groups ($F(\text{factor}) = \text{var.}(\text{factor})_a / \text{var.}(\text{factor})_b$).

For all groups, the variances (viz. the square of the standard deviation presented in table 3) attributable to the factor 'Type' (utterance type) increased from the measurement point 'mid schwa' to 'mid V', whereas the variance of the factor 'Speaker' and the 'Error' variance decreased (although not monotonically). In particular these last variances were of interest, because they expressed the variability between and within speakers that is not attributable to the vowel or the consonant in the utterance (utterance type). *F*-ratios showed that these within speaker variances ($F(\text{error}) = \text{var.}(\text{error})_a / \text{var.}(\text{error})_b$) were significantly larger for the NS children than for the adult women (all *F*s (391,383) > 1.46; $p < 0.05$). Moreover, the children with DAS showed significantly larger within speaker variances as compared to the NS children (all *F*s(495,391) > 1.70; $p < 0.05$). In contrast, the between speaker variances ($F(\text{speaker}) = \text{var.}(\text{speaker})_a / \text{var.}(\text{speaker})_b$) did not differ significantly between the groups (DAS/NS: all *F*s(8,5) < 5.61; NS/AW: all *F*s(5,5) < 1.97; ns.).

To summarize, it was found that the repeated utterances (error-variances) of the children were more variable than those of the adult women. The variability in children with DAS was even larger than in NS children. Although the variability within speakers differed significantly between the three groups (children with DAS have the highest variability within speakers, AW the lowest), the variability between speakers within groups did not differ significantly.

Table 3. Variability of second formant frequencies expressed in standard deviations in Hz (square root of variance) for each group: children with DAS, normally speaking (NS) children, and adult women (AW), subdivided into variability attributable to between- (Speaker) and to within-subject factors (Type and Error) (including percentage of the factor variance in proportion to the total variance). Significance of difference between groups was determined by calculating F ratios.

Group	Factor	Df	Mid schwa	End schwa	Cons	Onset trans	End trans	Mid V
DAS	Type	11	976.69	1456.25	2890.49	3979.17	4442.50	4454.49
	Speaker	8	2246.91	2008.11	1265.63	1291.21	959.95	934.97
	Error	495	361.46	390.85	288.55	318.27	305.03	201.22
NS	Type	11	1071.56	1623.27	2829.20	3936.58	4454.49	4770.10
	Speaker	5	1144.82	848.12	710.12	971.80	934.97	977.02
	Error	391	196.18	215.66	216.23	216.62	201.22	154.24
DAS / NS ¹	F (error)	495;391	3.38*	3.28*	1.78*	2.16*	2.30*	1.70*
	F(speaker)	8;5	3.85	5.61	3.18	1.77	1.05	1.09
AW	Type	11	610.00	748.59	2196.55	3442.79	3614.72	4257.84
	Speaker	5	1467.08	1384.73	506.29	707.38	740.71	729.75
	Error	383	162.40	164.77	157.58	119.27	134.44	113.98
NS / AW ¹	F (error)	391;383	1.46*	1.71*	1.88*	3.30*	2.24*	1.83*
	F(speaker)	5;5	1.64	2.67	1.97	1.89	1.59	1.79

Note. Significance * $p < 0.05$, after Bonferroni's correction; ¹ F ratio = largest variance / smallest variance.

Formant values and ratios

As a first result of the F2 patterns, it was found that the F2 values of the three groups differed significantly at all 6 measurement points (all Kruskal-Wallis Chi-square ($df=2$) > 18.2 ; $p < 0.05$). The F2 values measured in the utterances of the children with DAS were higher compared to NS children (significantly at each measurement point except consonant and mid vowel: Mann-Whitney $Z_s > 2.06$, $p < 0.02$), which in turn were higher than those of adult women (significantly at all measurement points: Mann-Whitney $Z_s > 2.11$, $p < 0.02$).

Difference in overall F2 values, i.e. some speakers had systematic higher or lower frequencies than others, complicate the comparison between speakers and groups. To facilitate a more valid comparison, as was stated in the 'method' section, F2 ratios were calculated as a method of speaker normalisation. For each speaker individually the formant values of utterances with /i/ were averaged and divided by the average formant values of utterances with /u/ (i/u-ratio). These ratios represent the distinction between the utterance types, i.e. the higher the ratio is above unity the more distinction there is between the utterance types at that particular measurement point. In figure 2 the i/u-ratios are presented throughout the utterance of the adult women (A), the NS children (B), and the children with DAS (C).

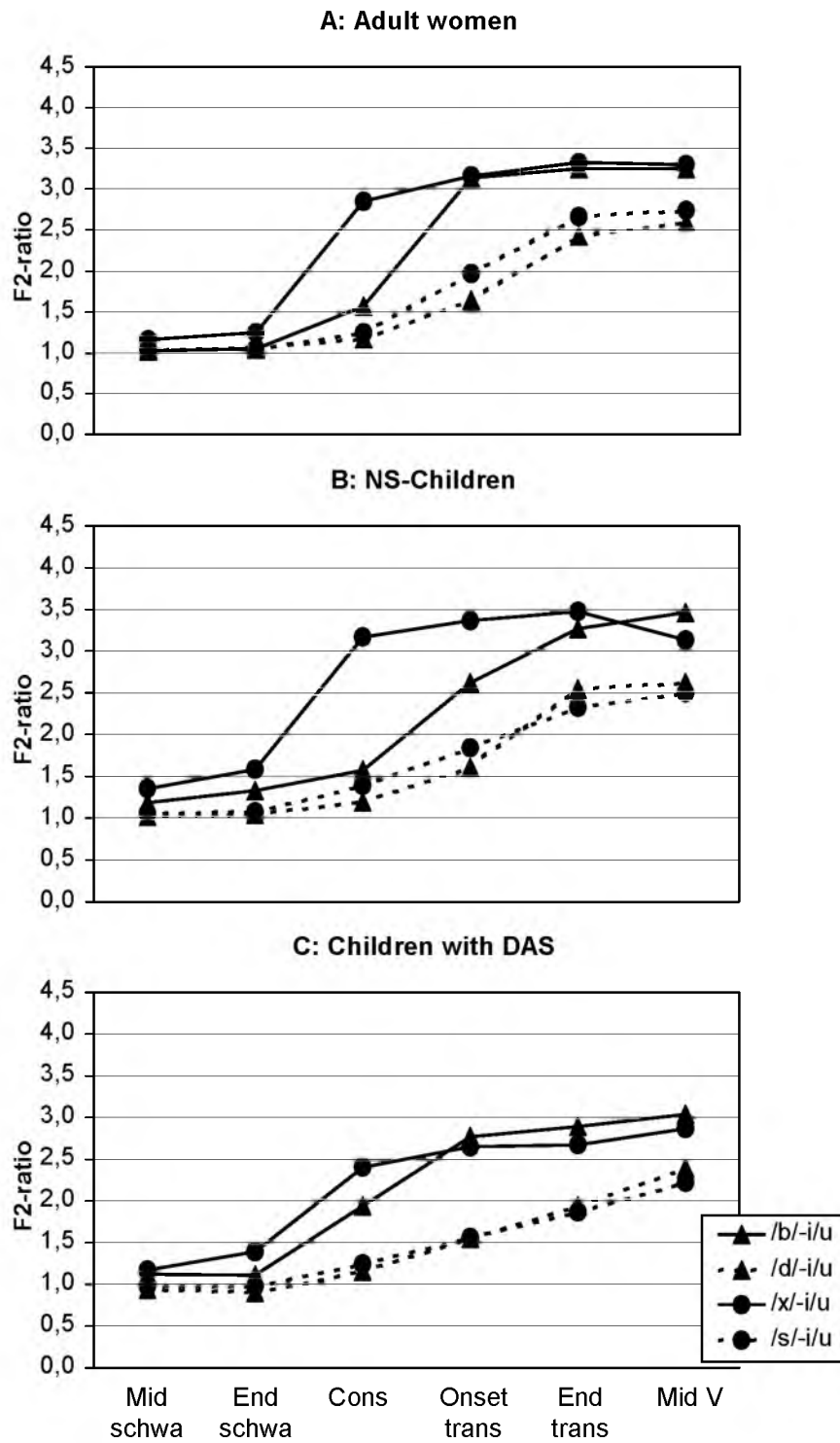


Figure 2. F2 ratios of Adult women (A) and NS children (B), and children with DAS (C).

High ratios were found at the location ‘mid V’, in all three groups. Of course, high ratios here were expected; they just demonstrate that the vowels used in this study are characterised by different F2’s. Children with DAS showed smaller i/u-ratios as compared to NS children and adult women ($F(2,79)=4.23$; $p<0.05$; post-hoc LSD DAS versus NS: mean difference = -0.394, $p<0.025$; NS versus AW: mean difference = -0.040, ns.), which indicated that the distinction between the /i/ and /u/ was not as large in children with DAS as compared to the other groups. The more interesting part is formed by the ratios earlier in the utterance (all measurement points before Mid V), since these ratios indicate an effect of the upcoming vowel, i.e. anticipatory coarticulation of the vowel on the preceding schwa (intersyllabic coarticulation) and consonant (intrasyllabic coarticulation). Going from right to left, diminishing ratios can be observed in all figures (i.e. smaller ratios in the schwa than in the consonant), so there seemed to be diminishing effects from the upcoming vowel on the F2 values in the preceding consonant and schwa.

The ratio figures suggest that the strength of this anticipatory coarticulation was different for the different consonants (the dashed lines in figure 2 are closer to unity than the solid lines): the utterances with /d/ and /s/ showed weaker coarticulation than the utterances with /x/ and /b/ (in particular the /x/-utterances). An analysis of variance showed that the factor Consonant had a significant effect on the ratios of all measurement points in adult women and NS children (AW: all F_s (3,15)>5.62; NS children: all F_s (3,15)> 7.59; $p<0.01$), and in children with DAS on all measurement points but two (end schwa and mid V). However, the effects were stronger in adult women and NS children than in children with DAS (NS: η^2 : 0.60-0.89; AW: η^2 : 0.55-0.89; DAS: η^2 : 0.30-0.67). A striking feature in the figures of Adult women (2A) and NS children (2B) was the trajectory of the /x/-i/u ratio in the schwa and the consonant. These high /x/-i/u ratios were mainly caused by low F2 frequency in the /xu/-utterances, which was lower than all other /Cu/-utterances.

In order to evaluate the significance of the influence of the vowel on the preceding sounds (i.e. testing the null hypothesis that F2 ratios equal 1), one-sample t -tests were conducted for each speaker group separately (in order not to violate homoscedasticity) and each consonant separately. Results of these analyses are displayed in table 4.

The results in table 4 corroborate the patterns in the figures. In the consonant the F2 ratios are higher than 1 for all groups and consonant contexts (except for /b/-utterances in NS children), indicating intrasyllabic coarticulation. The F2 ratios in the schwa of adult women and NS-children were significantly higher than 1 (indicating intersyllabic coarticulation) for all utterances except the /b/-utterances in adult women. Comparing both groups, it was found that the ratios were higher in the schwa of NS children as compared to adult women (mid schwa $F(1,46)=4.67$; end schwa $F(1,46)=5.13$; $p<0.05$). This means that the intersyllabic coarticulation in NS children was stronger than in adult women. In children with DAS the t -testvalues were not significantly higher than 1 in the schwa, which means that no significant intersyllabic coarticulation effect of the vowel on the preceding schwa could be found in DAS. However, in the comparison of the ratios of the children with DAS to those of the NS children no group effect was found in the schwa and consonant, but only in the vowel (transition end $F(1,56)=6.23$; mid vowel $F(1,56)=4.42$; $p<0.05$). Thus, an explanation for the lack of significant results in t -test

values in the schwa of children with DAS might lay in the high variability in DAS rather than in the absence of intersyllabic coarticulation.

Table 4. One sample *t*-test for each group (children with DAS, normally speaking (NS) children, and adult women (AW)) and each consonant separately.

Group	Cons	Mid schwa		End schwa		Cons		Onset trans		End trans		Mid V	
		df	<i>t</i>	df	<i>t</i>	df	<i>t</i>	df	<i>t</i>	df	<i>T</i>	df	<i>t</i>
AW	/b/	5	0.56	5	0.87	4	4.36*	4	9.15***	5	10.10***	5	24.14***
	/d/	5	2.65*	5	3.26*	2	4.87*	4	15.03***	4	6.07**	5	12.88***
	/x/	5	4.77**	5	4.82**	5	8.42***	5	30.23***	5	12.17***	5	14.83***
	/s/	5	3.92*	5	4.30**	5	6.27**	4	5.16**	5	9.66***	5	14.09***
NS	/b/	5	4.06**	5	5.22**	4	1.87	5	6.48***	5	5.82**	5	8.17***
	/d/	5	5.04**	5	2.83*	5	8.21***	5	7.59***	5	12.20***	5	9.53***
	/x/	5	4.84**	5	4.77**	5	10.13***	5	10.28***	5	8.02***	5	10.42***
	/s/	5	2.85*	5	3.58*	5	5.37**	5	3.51*	5	9.48***	5	9.47***
DAS	/b/	7	1.20	5	1.26	6	3.77**	7	4.96**	7	7.86***	7	6.60***
	/d/	7	-0.06	7	-0.02	6	4.72**	6	2.91*	6	4.19**	7	5.56***
	/x/	8	2.01	8	2.28*	7	7.27***	8	6.99***	8	9.98***	8	8.16***
	/s/	8	1.32	8	1.22	7	5.08***	7	2.34*	8	3.24**	8	5.09***

Note. Significance* $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$.

To summarize the ratio patterns, in all three groups of speakers intrasyllabic coarticulation was found, the strength of which was dependent on the consonant (stronger effects in /b/- and /x/- compared to /s/- and /d/-utterances). Moreover, NS children and adult women showed intersyllabic coarticulation that was stronger in NS children than in adult women. No significant intersyllabic coarticulation could be found in children with DAS, which might be due to the high variability in DAS.

Individual speakers

As was discussed before, on the basis of the variances between and within speakers, children were more variable as compared to adult women. Furthermore, the children with DAS displayed more variability than the NS children did. Although no significantly larger between subject variances were found in the comparison of the NS children to the adult women, nor in the comparison of the children with DAS to the NS children, the individual ratio patterns showed different patterns among children, especially among the children with DAS. Therefore, the ratio patterns of the individual children are presented in figure 3 (A-O). In order to test the significance of the distinction between the /i/ and /u/-utterances Mann-Whitney tests were conducted on the F₂ values for each child and consonant separately.⁷ Table 5 shows the

⁷ One sample *t*-tests on the F₂ ratios could not be conducted, because there was only one ratio-value per child per consonant.

consonant contexts in which significant differences between the F2 values of /i/- versus /u/- utterances were found, in the schwa (mid schwa or end schwa) and/or in the consonant.

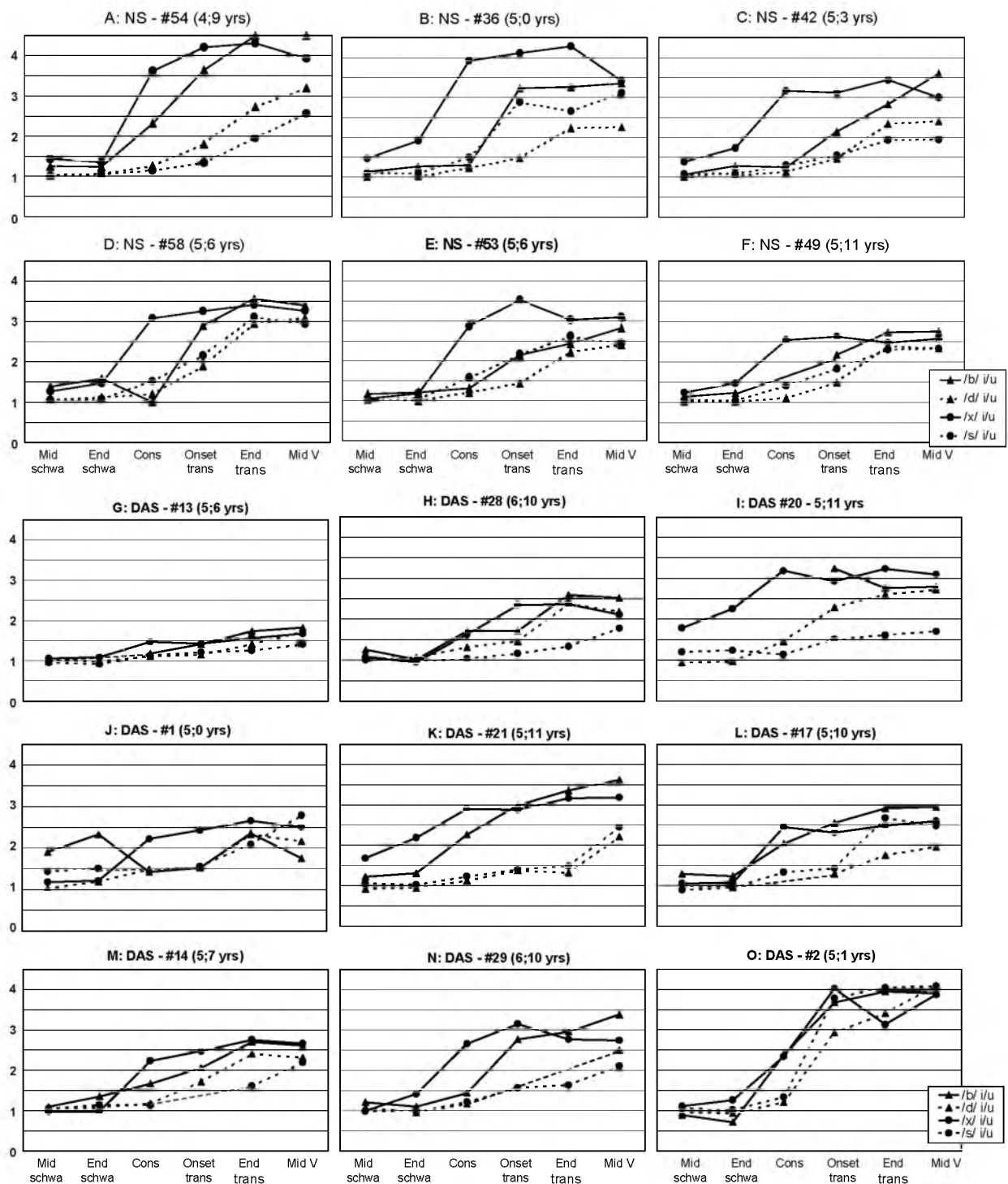
Table 5 shows that almost all NS children had significant intrasyllabic coarticulation effects of the upcoming vowel on the consonant. Only in the /b/, we could not find significance since not enough valid values were available to compute the statistics. Every NS child did show a significant difference between /i/ and /u/-utterances measured in the schwa (intersyllabic coarticulation) in the /x/-utterances. Four of them reached significance in the /b/-utterances. The utterances with /d/ showed the least intersyllabic coarticulation; only two out of six children showed significant distinction between /i/- and /u/-utterances determined in the schwa.

Table 5. Vowel coarticulation effects for preceding schwa and consonant as assessed by the differences between the F2 values of /i/-utterances and the /u/-utterances. A '+' indicates this difference was significant (determined by Mann-Whitney tests for each child and each utterance type (x, b, s, d) separately).

Group	Child	/x/-utterance		/b/-utterance		/s/-utterance		/d/-utterance	
		Schwa	Cons	Schwa	Cons	Schwa	Cons	Schwa	Cons
NS	#54	+			+				+
	#36	+	+	+	1)	+	+		+
	#42	+	+		1)	+	+	+	+
	#53	+	+	+			+		+
	#58	+	+	+	1)	+	+	+	+
	#49	+	+	+	1)	+	+		
DAS	#13			1)					
	#28	1)	1)					+	
	#20	+	1)	1)	1)		1)		
	#1		+			+	1)		+
	#21	+	+	+	+		+	+	+
	#17		+	+	1)		+		1)
	#14		+	+	+	1)			+
	#29	+	+	1)			+	1)	+
	#2	+	+		+		+		

Note. 1) = not enough valid values to compute the statistic.

The ratio figures of the NS children (figure 3A-F) show that although the individual children differed in extent of coarticulation of the vowel (there appears to be a tendency for younger children displaying higher ratios) the pattern among the individual children was highly similar.



Note. NS children are ordered according to age; children with DAS according to similar patterns.

Figure 3. Ratio-figures of the individual NS children (A-F) and the children with DAS (G-O).

The ratio figures (figure 3G-O) and statistical results (table 5) of the children with DAS display different and inconsistent patterns.⁸ Some children with DAS showed very flat ratio curves, with very low ratios and hardly any differentiation in succeeding segments. Child #13 (figure 3G) is a striking example of this. The ratios were very low throughout the utterance. A similar pattern was found in child #28 (figure 3H), however the patterns showed slightly more differentiation in succeeding segments. Child #20 (figure 3I) also showed a pattern of flat curves, although the ratios are much higher. Remarkable are the high ratios in the schwa of the /x/-utterances, which displayed strong intersyllabic coarticulation. Child #1 (figure 3J) also exhibited rather flat curves. Still, intersyllabic coarticulation was found in the /s/-utterances and intrasyllabic coarticulation in the /x/- and /d/-utterances.

Child #21 (figure 3K) shows high ratios in the schwa (intersyllabic coarticulation) for the /x/- and /b/-utterances. In the utterances with /d/ and /s/ the transition to the vowel was made late in the utterance, just before vowel midpoint. Still, intrasyllabic coarticulation was significant in all utterance types.

Other children, Child #17, #14, and #29 (figure 3L-N) show more differentiation between succeeding segments, with weak intersyllabic coarticulation and strong intrasyllabic coarticulation, especially in the /x/-utterances.

Finally, the curves of child #2 (figure 3O) are divergent from the patterns of all other children. This figure shows very high ratios in the vowel that started either at transition onset (in the utterances with /s/ and /d/) or in the consonant (in the utterances with /x/ and /b/); even higher than was found in most NS children. Strong intrasyllabic coarticulation was found in the utterances with /x/, /b/, and /s/, but intersyllabic coarticulation was only found in the /x/-utterance. The ratios in the utterance with /b/ are even below unity.

To summarize the ratio results of the individual children, the patterns of the individual NS children showed more uniformity than those of the children with DAS. The utterances of children with DAS showed smaller F2 ratios throughout the total utterance (except for child #2, figure 3O) as compared to NS children, which represent less differentiation between the different utterances, and rather flat curves that indicate reduced acoustic differentiation between the successive speech segments.

DISCUSSION

The question for this study was, can *weaker* or *stronger* coarticulation patterns be demonstrated in DAS as compared to NS children? And, if so, are these effects restricted to the syllable (intrasyllabic) or do they cross the syllable boundary (intersyllabic)? These questions were addressed by using formant frequency measurements. The first result was, that the variability of the formant values in the group of children with DAS was larger than in the NS children and in the adult women (see table 3). On the other hand, the variances between speakers within groups did not differ significantly across the three groups. As a second result, although both NS children

⁸ The statistical results have to be taken with some reservation, because due to the high intrasubject variability in the children with DAS the power of the statistical tests will be lower as compared to the NS children, which might lead to a smaller number of significant results.

and adult women displayed highly similar patterns of F2 ratios, NS children exhibited intersyllabic coarticulation in all utterances whereas adult women did not show intersyllabic coarticulation in /b/-utterances. In contrast to the NS children and adults, the children with DAS as a group made less distinction between the vowels (F2 ratios were lower at mid V, figure 2C). Furthermore, children with DAS produced idiosyncratic ratio patterns and therefore no significant intersyllabic coarticulation was found in this group of children.

Thus, the main result was on the whole less distinction between the vowels and more variability resulting in idiosyncratic patterns in children with DAS as compared to NS children and adult women. We will further elaborate the issue of normal and pathological speech and speech development below.

Normally speaking children versus adult women

Studies on the normal development of coarticulation show diverse results. Two opposite views are discussed here. The first view is represented by Nittrouer and colleagues (1989; 1993; 1996) and Siren and Wilcox (1995). In a series of coarticulation experiments Nittrouer et al. showed that in normal speech development a decrease in coarticulation between syllables (intersyllabic coarticulation: effect of V on preceding [ə]) was found earlier in development than a decrease in coarticulation within syllables (intrasyllabic coarticulation: effect of V on preceding C). Nittrouer et al. concluded that in development the phonetic segment is the endpoint rather than the starting point of development.

On the other hand, some researchers propagate the opposite view according to which during development the skill of syllable cohesion is learned after the child has mastered the articulation of the individual segments. From this perspective, the speech of children should reveal less evidence of coarticulation within and between syllables than the speech of adults. Evidence supporting this view comes from studies showing that anticipatory coarticulation (lingual and labial gestures) is more apparent in adults' than in children's utterances, both acoustically and perceptually (Kent & Rosenbek, 1983; Sereno, Baum, Marean, & Lieberman, 1987; Sereno & Lieberman, 1987). Sussman, Duder, Dalston, & Cacciatore (1999) found both weaker and stronger coarticulation effects in a single female child depending on the articulators involved. The divergent results found in studies investigating the development of coarticulation might be due to differences in methods and utterances used to study coarticulation.

In the present study the utterances of NS children exhibited stronger intersyllabic coarticulation effects than adult women's utterances as was shown in figure 2A and 2B (and in the statistical results). Intrasyllabic coarticulation was found in all utterances, but was weaker in utterances with alveolar consonants (/d/ and /s/) than in the utterances with /b/ or /x/. These results support the view of Nittrouer et al. that *more* coarticulation is found during development compared to adult speech. Furthermore, more variance was found in the repeated utterances of NS children as compared to those of adult women, which indicates inconsistency. This inconsistency can be interpreted as immaturity of the children's speech production system, in which the speech production process is not fully automated.

In summary, the results of this study support the idea that normal speech development leads to more consistency in repeated utterances and to smaller intersyllabic coarticulation effects. With

this in mind the coarticulatory effects as found in the utterances of children with DAS will be re-examined.

Developmental apraxia of speech

Like developmental studies on coarticulation, studies on coarticulation in acquired apraxia also reported divergent results (Dogil et al., 1996; Katz & Baum, 1987; Katz, 1988; Southwood, Dagenais, Garcia, & Sutphin, 1996; Whiteside & Varley, 1998; Ziegler & Von Cramon, 1985; 1986). The conclusions of these studies varied from an absence of coarticulatory cohesion due to a lack of automation (Whiteside & Varley, 1998) to a preservation of coarticulation depending on articulators involved and speaking rate (Southwood et al. (1996) found distorted labial coarticulation at all speaking rates and distorted lingual coarticulation at fast speaking rate). Therefore, the design of the present study included both children with DAS and control groups (normally speaking children and adult women), in order to overcome interpretation problems due to differences in speech tasks and material.

In the comparison between children with DAS and NS children, as a first result, significantly larger variances in the repeated utterances were found in children with DAS as compared to NS children. This was not surprising, since high variability is often found in speech disorders (McCabe, Rosenthal, & McLeod, 1998). These larger variances, indicating inconsistency, can be interpreted as less mature automation. An explanation for the small number of significant differences in variance between speakers in children with DAS as compared to NS children could be found in the fact that only the phonemically correct utterances were used in the analysis. More variability might have been found if all utterances were considered for analysis.

As a second result, the average F2 values of children with DAS were higher than those of NS children. As shown in table 2, not only F2 but also F1 values were, on average, higher in the DAS than in the NS group (this was not statistically analysed). Since the F2 is determined by the size of the front cavity (which is influenced by both the shape of the back of the tongue and liprounding) and the F1 is related to both jaw opening (F1 is higher when jaw opening is larger) and length of vocal tract (inversely proportional) an explanation for these group differences in F2 values does not seem to be straightforward (Zemlin, 1988). Furthermore, all three target vowels displayed such a group difference, which might be explained on the basis of differences in general size factor (due to anatomical differences such as growth rate) rather than behavioural aspects such as lip protrusion. However, this last explanation does not seem logical since there is no independent evidence for this and, moreover, the two groups of children were matched with respect to age. Thus, no convincing explanation was found for the group differences in F2 values.

Thirdly, average F2 ratios of children with DAS displayed less differentiation between utterance types, especially measured in the vowel, compared to the average F2 ratios of NS children. A reduction in vowel distinction (i.e. children with DAS do not make as much distinction between the different vowels) is expected to lead to less distinction between the utterance types measured earlier in the utterance, i.e. in the preceding consonant and schwa, which indicates a reduction of coarticulation. After all, the smaller the distinction between vowels, the less this effect can spread backwards to the preceding phonemes. The results of the

individual children, however, suggest that a reduction of vowel distinction does not necessarily imply reduced coarticulation. The NS child #49 (figure 3F; table 5) displayed small distinction in the vowel, yet significant inter- and intrasyllabic coarticulation. And vice versa, an increase in vowel distinction does not necessarily lead to larger coarticulation effects; the DAS child #2 (see figure 3O and table 5) displayed a large distinction in the vowel, yet only significant intersyllabic coarticulation in the /x/-utterances. Because of this, the children were considered individually. These results showed inconsistent patterns among children with DAS. Some children displayed very flat curves (child #13, #28, #20, #1) with hardly any intra- and intersyllabic coarticulation. Another child (Child #21) showed large coarticulation effects in the /x/ and /b/ utterances, but very small coarticulation effects in the utterances with /d/ and /s/. Other children (child #17, #14, #29, and #2), however, showed more segmented speech with weak intersyllabic coarticulation. These divergent results contrasted with the consistent intrasyllabic coarticulation in the NS children.

Finally, a possible weaker intersyllabic coarticulation effect in children with DAS may be associated with difficulty in grading syllable stress (Shriberg et al., 1997). If they produced the schwa as in a strong rather than as in a weak syllable, this strong syllable may be less ‘susceptible’ to influence by the upcoming vowel.

In conclusion, the F2 values in the utterances of the children with DAS can be characterised by (1) large variability, (2) higher F2 values, (3) lack of distinctiveness between the utterances, especially shown in the F2 ratios measured at vowel midpoint, (4) a smaller number of inter- and intrasyllabic coarticulation effects (although this might be due to lower statistical power), and (5) idiosyncratic patterns. The first result, larger variability was also found in the NS children as compared to the adult women. This can be interpreted as ‘younger speech’ characterised by less automation. Results of a follow-up study 1,3 years later might further test this hypothesis. The divergent patterns of coarticulation that were found in the utterances of the individual children with DAS, however, indicate that it is not merely a delay in speech. Each child has its own motor pattern to cope with the problems in the speech production process.

Homorganic versus heterorganic

Hardcastle and Edwards (1992) showed that the tongue could be considered as two separate articulators: tongue body and tongue tip/blade. The production of the velar fricative [x] involves the tongue body and the tongue tip is used for the alveolar [s]. Vowels are typically produced by the tongue body. As a consequence of this, as far as coarticulation is concerned, the critical difference between the different utterances is the number of articulators involved. Thus, [əxu] and [əxi] can be considered homorganic (consonant and vowel share the tongue body as articulator). On the other hand, the sequences [əsu] and [əsi] can be considered heterorganic (both tongue-articulators are involved: tongue tip/blade for [s] and tongue body for [u] or [i]). A similar distinction between homorganic and heterorganic occurs in the utterances [əbu] and [əbi]. In the homorganic [əbu] consonant and vowel share one main articulator, the lips, for closing and rounding whereas in the heterorganic [əbi] lip rounding is not necessary. Note that [əbu] is a homorganic sequence with respect to a different articulator (lips) than [əxu] (tongue body). In the

utterances [ədu] and [ədi] the consonant and the vowel do not share the main articulators tongue body or lips.

A closer inspection of the F2 data yields additional, interesting results related to this homorganic and heterorganic distinction. The utterances with the alveolar consonants /d/ and /s/ showed smaller coarticulation effects, especially intersyllabic, than the utterances with labial /b/ and velar /x/ consonants. In the /b/-utterances this is mainly due to the influence of the vowel /u/ on the preceding sounds. This finding is compatible with the findings of Fowler and Brancazio (2000), who found differences in coarticulation resistance of consonants, in which the consonant /b/, /v/, and /g/ showed less resistance than the consonants /d, ð, z, ʒ/. If we think of coarticulation as competition for the same articulators, we might infer that consonant-vowel sequences that share the same articulator, i.e. are homorganic (e.g. lips in [bu]), cause articulatory movements to overlap more, resulting in stronger coarticulation effects, than consonant-vowel sequences that do not share the same articulator, i.e. are heterorganic (e.g. [si]). Nitttrouer (1993) compared coarticulation in /k/ and /t/ contexts and concluded from stronger intersyllabic coarticulation in /k/ than in /t/ that ‘...the tongue was apparently freer to move in anticipation of the upcoming vowel in the /k/ context than in /t/ context.’ (p.967). Our data corroborated this difference between velar and alveolar for all groups of speakers. Thus, the factor homorganic versus heterorganic plays a consistent role in the extent in which coarticulation occurs.

The stronger intersyllabic coarticulation effects in NS children as compared to adult women for the utterance /əbu/ suggest that the children who participated in this study had not yet developed an adult coarticulation pattern. Furthermore, it suggests that homorganic articulation patterns mature later than heterorganic articulations. The results of a second measurement (1,3 years later), to be presented in a following publication will reveal more data on coarticulation patterns in normal development.

Methodological issues

A methodological concern of this study bears upon the reliability and validity of formant analyses. It is well documented that formant measurements in women and children’s speech is problematic (e.g. Bennett, 1981). Therefore, in the present study several precautions were taken to improve reliability and validity of the measurements. First, unequivocal criteria were established to segment the acoustic signals into speech sounds. In particular for the most relevant segments (schwa, consonant and vowel), onsets and offsets were clearly demarcated. In order to assess reliability two researchers segmented part of the speech material. It turned out that the inter-observer reliability was quite satisfactory. Second, two sources of information were used to determine formants. For each speaker several tokens of more than one utterance type were thoroughly inspected with respect to the correspondence between the Fast Fourier spectrogram and the LPC-determined formant values on the basis of the formant values plotted overlaid on spectrograms. From these inspections ranges of formant values for that particular speaker were assessed, which were later used to remove outliers. Moreover, *all* spectrograms and formant traces were visually inspected for erratic patterns. We are convinced having optimised the measurements by adopting such a speaker-adapted procedure. The low variability and low dropout rates of the utterances spoken by the adult women support the success of this

optimisation. Higher variances were found for the NS children, and even higher for the children with DAS. Although it is hard to distinguish increased variability in productions from increased measurement error, the thorough procedure used seems to warrant our conclusions. Besides this, as a consequence of this higher variability in children the power of statistical tests is smaller, which led to a smaller number of significant differences in vowels in table 5. Thus, seemingly large coarticulation effects as appears in the ratio figures of the individual children did not always reach significance in table 5.

An issue for implementation of these procedures for clinical assessment is the fact that the procedure described above is very time consuming. This forms a serious impediment for large-scale implementation. In a future study we aim at developing a more automatic measurement procedure.

A separate problem is the measurement of the second formant frequencies in the consonants, particularly the plosives. LPC-measurements in the plosive bursts frequently resulted in large formant bandwidths. Because of the strict criteria we used, many of these resulted in missing values. Much less problems were encountered in the fricatives, which corresponds to the results reported by Nittrouer et al. (1993; 1996). Currently, the spectral moments of these same utterances are measured (Maassen, Nijland, & Van der Meulen, 1999). These will be compared to the formant frequencies.

Besides the method described in this study to show coarticulation, the method of determination of locus equations (described for example in Sussman et al., 2000) was also executed. However, since the speech material was not constructed for this purpose, the small number of different vowels (only three instead of ten used by Sussman et al., 2000) did not result in reliable slope calculation. Therefore, the results could not be interpreted reliably and were not shown here.

Final conclusion

To conclude, acoustical analysis of the speech of children with DAS, as was done in the present study, reveals phonetic deviancies as compared to normal speech that cannot be revealed by phonetic transcription or perceptual judgement. This helps to clarify the underlying problem of children with DAS. We realise that studying only children with DAS and no children with other speech disorders we cannot draw any conclusions as to the specificity of the findings for the speech disorder DAS. However, the results do provide information about DAS. The patterns can be interpreted as a problem in the automation of speech production. The results of this study strongly suggest that speech production of children with DAS is not only delayed but also deviant. These hypotheses will be further tested in a follow-up study 1;3 years later.

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CHAPTER THREE

Planning of syllables

Nijland, L., Maassen, B., Van der Meulen, S.J., Gabreëls, F., Kraaimaat, F.W., & Schreuder, R. (2003). Planning of syllables in children with Developmental apraxia of speech. *Clinical Linguistics and Phonetics*, 7 (1), 1-24.

ABSTRACT¹

The aim of the present study was to investigate whether children with developmental apraxia of speech (DAS) show a deficit in planning syllables in speech production. Six children with DAS and six normally speaking (NS) children produced high- and low frequency of occurrence syllable utterances, in which the syllable structure was systematically manipulated in an otherwise unchanging phoneme sequence. Anticipatory coarticulation, using second formant trajectories, and durational structure were analysed. The results showed stronger coarticulation in the children with DAS when compared to the normally speaking children, but in contrast to our expectations, in neither group was a systematic effect of syllable structure on the second format trajectory found. Effects of syllable structure did emerge for durational structure in that durational adjustments were found in the segments of the second syllable. These adjustments were less systematic in children with DAS when compared to normally speaking children. Furthermore, at the prosodic level, normally speaking children showed metrical contrasts that were not realized by the children with DAS. The latter results are interpreted as evidence for a problem in the planning of syllables in speech production of children with DAS, in particular concerning prosodic aspects, which is discussed in relation to the automation of speech production.

INTRODUCTION

In this study we investigated the use of syllables in children with developmental apraxia of speech (DAS). DAS is a speech disorder that interferes with the ability to produce intelligible speech due to an impairment in sequencing speech sounds, syllables and words. The speech of children with DAS is, therefore, highly unintelligible as a result of the large number of segmental errors, especially consonantal errors, (contextual) substitutions and omissions. Inconsistency of errors and groping (articulatory searching behaviour) are typical of DAS. Also, errors and distortions in vowel production have been reported (Pollock & Hall, 1991; Walton & Pollock, 1993). In addition to studies of segmental error productions, research on prosody profiles has documented inappropriate stress patterns in children with DAS (Shriberg, Aram, & Kwiatkowski, 1997). It was found that children with DAS made less distinction between stressed and unstressed segments when compared to normally speaking (NS) children.

Although it is still unclear at precisely what level of speech production processing the underlying deficit that causes DAS is localized, several production models suggest that the origin can be found somewhere in the transition from a phonological code into articulo-motor output, that is, in phonetic planning, motor programming, or motor execution (e.g., Dodd, 1995; Hall, Jordan, & Robin, 1993; Ozanne, 1995; Van der Merwe, 1997; Velleman & Strand, 1994). Velleman and Strand (1994) suggested that children with DAS are impaired in their ability to generate and utilize frames. Thus, children with DAS ‘might “have” appropriate phonological (or syntactic) elements but be unable to organize them into an appropriate cognitive hierarchy’ (p. 120). They argue that although children with DAS display a large number of segmental errors, the crucial supposition is that the size of the unit underlying these errors is not the single segment

¹ The journal in which this chapter is published uses British-English spelling therefore this chapter has British-English spelling rather than American-English.

itself, but the syllable in which the segment is embedded. An example of this is the often-reported inconsistency of errors produced by children with DAS (Thoonen, Maassen, Gabreëls, & Schreuder, 1994).

There are several characteristics of apraxic speech that support an interpretation in which syllable context plays a predominant role in speech production. Examples are: deficits in timing and co-ordination that have been reported in voice-onset-time studies (e.g., Kent & Rosenbek, 1983; Ziegler & Von Cramon, 1986a); and delayed transitions and problems in phasing the articulatory movements in apraxia of speech (e.g., Whiteside & Varley, 1998; Ziegler & Von Cramon, 1985; Ziegler & Von Cramon, 1986b). These types of errors are more easily described in terms of articulatory syllabic context influences, than in terms of phoneme selection and sequencing. Furthermore, recent results of Marquardt, Sussman, Snow and Jacks (2002) suggested a breakdown in the ability of children with DAS to perceive 'syllableness' and to access and compare syllable presentations with regard to position and structure.

Likewise, syllabic context plays a role in the planning and programming of speech, i.e., in the transition from a phonologic representation to the motor programme (Ozanne, 1995, cf. Levelt, 1989). This transition comprises the collection of spatial and temporal goals of the articulatory movements for speech sound productions, conceptualized as gestures by Browman and Goldstein (1997), from a sensorimotor memory and adapting these to the surrounding sounds. The result is a motor plan, or gestural score (Browman & Goldstein, 1997), which is subsequently translated into a motor programme by specifying the exact movements of the speech musculature, i.e., muscle tone, rate, direction and range of movements (Van der Merwe, 1997). The present study investigated whether children with DAS show a deficit in using the syllable as a planning or programming unit.

Previously we stated that in normal speech the coherence of the spatial, temporal and scaling aspects of gestures is preserved by using syllable-sized gestural scores (Löfqvist, 1990). This means that within the syllable, a particular speech sound is dependent on the characteristics of the adjacent speech sounds, which induce coarticulation (e.g., the articulation of the [t] in 'tool' exhibits liprounding under the influence of the following, rounded vowel [u]) and durational adjustment. As a consequence of this syllabic organization it is predicted that durational adjustment and coarticulation within the syllable (intra-syllabic coarticulation) is stronger than between syllables (inter-syllabic coarticulation) (Browman & Goldstein, 1997; Levelt, Roelofs, & Meyer, 1999). Evidence for deficient planning of syllables in DAS would, therefore, come from particular deviant coarticulation and durational patterns between and within syllables.

In previous research, we investigated the coarticulation patterns of children with DAS, and compared them to normally speaking children and adult women (Boers, Maassen, & Van der Meulen, 1998; Nijland, Maassen, Van der Meulen, & Bellaar, 1999; Nijland et al., 2002). The results of these studies indicated that the speech of the children with DAS showed deviant coarticulation patterns as compared to the normally speaking children, with overall weaker coarticulation and less spatial (articulatory) distinction between vowels. The individual patterns of the children with DAS were characterized by idiosyncrasies, which were interpreted as indications of deviance. Besides these deviant patterns, the children with DAS, when compared to normally speaking adults and children, also displayed the highest variability in repeated utterances. From

these results it was concluded that children with DAS showed a lesser degree of coarticulatory cohesion.

In the present study, coarticulatory cohesion is further investigated; in particular, coarticulation in DAS is compared to normal speech. We introduce a more rigorous experimental manipulation of the speech material in order to determine whether the deviant coarticulation patterns and durational structure reflect a disability in planning syllables. Because gestural scores are hypothesized to be organized in syllables, we predict a strong effect of syllable structure on coarticulatory cohesion and durational structure. For this reason, children produced utterances in which syllable structure was varied, while the phoneme sequence did not change. An example in English would be the phrase ‘ice cream’ versus ‘I scream’, in which the syllable boundary follows the /s/-sound in the first phrase and precedes the /s/ in the latter.² Notice that the sequence of phonemes is identical in both utterances.

Coarticulatory cohesion (i.e., the influence of surrounding phonemes) within and between syllables, and the durational pattern in the utterances of normally speaking children and children with DAS, is substantiated with formant and durational measurements (Lee, Potamianos, & Narayanan, 1999; Nittrouer, 1993; Nittrouer, Studdert-Kennedy, & Neely, 1996; Ziegler & Von Cramon, 1985; 1986a; 1986b). Because it was not known beforehand what the effect of the manipulation of syllable structure would be for normally speaking children, the speech material was constructed so that a particular coarticulation effect could occur within a syllable and across a syllable boundary. We made use of the coarticulation effect of a vowel on the preceding consonants. Under the hypothesis (that normal speech is organized in syllable-length gestural scores) it is expected that the influence of the vowel on the preceding consonant is stronger and more robust if that consonant belongs to the same syllable, than if that consonant belongs to a different one, namely the preceding syllable. Such a syllable structure effect is expected to appear both spectrally (in formant measures) and temporally (in durational measures). Thus, in the English example above, the coarticulation of the vowel [i] (in ‘-cream’) on the [s] would be stronger in the phrase ‘I scream’ than in the phrase ‘ice cream’. Such an effect cannot be attributed to phonemic differences because the phonemic sequence is identical in both cases; instead, it must be caused by differences in syllable structure.

Such a syllable-internal dependence of gestures made Levelt et al. (1999: 5) suggest that ‘a speaker has access to a repository of gestural scores for the frequently used syllables of the language’, i.e., the syllabary (following the suggestion of Crompton, 1982: 32, in Levelt et al., 1999). The function of these stored patterns is to relieve the production process of the task of computing the motor plan for the production of a particular syllable time and again. Instead, after an acquisition period, the articulatory patterns of frequently uttered syllables are stored, and can be retrieved on demand. Thus conceived, the syllabary can be interpreted as a mechanism for automation (Varley & Whiteside, 2001; Ziegler, 2001). Syllables that do not occur in the language are not stored in the syllabary. Based on this conceptualization, a difference between intra- and inter-syllabic coarticulation is expected, particularly in high-frequency syllables, which are

² In this example, the syllable structure is equal to the word structure, since it concerns a one-syllable word. We will use the term ‘syllable structure’ and ‘syllable boundary’ henceforth, because the influence of the syllable is important in this study.

supposed to be stored in the syllabary. To evaluate this hypothesis further, the speech material of the present study was constructed with high-frequency syllables as well as low-frequency syllables.

Knowing the effects of syllable structure and syllable frequency for normally speaking children, the next question will be: do children with DAS produce similar or different effects? Problems in planning syllables are predicted to emerge in DAS as *deviant* coarticulatory and durational patterns dependent on syllable structure. Thus, if we find that children with DAS produce a difference between intra- and inter-syllabic coarticulation strength and durational structure, similar to the difference of normally speaking children, then we must conclude that the deficit in DAS is not particularly located in the planning of syllables. On the other hand, if children with DAS do not show a syllable structure effect like that of normally speaking children, then this finding would be interpreted as a disturbance in children with DAS with respect to the ability to plan syllables, possibly due to deficient storage and/or accessibility of the syllabary. This latter interpretation would be further supported if a smaller effect of syllable frequency were found in children with DAS when compared to normally speaking children. Thus, the question of this study whether children with DAS show a deficit in planning syllables (including the use of a syllabary) and is investigated by manipulating syllable structure as well as syllable frequency.

METHOD

Participants

Data were collected from six children with DAS and six normally speaking children. These individuals were randomly selected from a total number of 19 children with DAS (14 boys and 5 girls; age range: 4;11–6;10 yrs) and 19 normally speaking (NS) children (matched for sex, age and dialect). All children were native speakers of Dutch (the reason for analysing only six children with DAS and six normally speaking children is that the detailed acoustic analyses presented in this study are extremely time consuming and therefore could not be conducted on all 38 children).

The children with DAS were ‘clear’ cases selected from special schools for children with speech and language disorders. The selection procedure was as follows: speech-language pathologists referred 70 children with suspected DAS to the authors. These children were further evaluated on the basis of the following tasks: spontaneous speech, repetitive imitations of phonemes, words and brief phrases, repetitions of nonsense words, and a diadochokinetic task (collected by the school speech-language pathologist). The following criteria were adopted: exhibiting many phonemic errors despite a complete phoneme repertoire (as compiled in the imitation tasks), high-frequency consonant substitutions (and omissions in clusters), phoneme and syllable sequencing difficulties which emerge in repetitions and in groping behaviour, inconsistent error patterns in repetitions of words and phrases, and inability to produce complex phonemic sequences (Hall et al., 1993; Thoonen, Maassen, Wit, Gabreëls, & Schreuder, 1996). Additional selection criteria included: (at least) average non-verbal intelligence level, no problems with hearing or with language comprehension, no organic disorders in the orofacial area, and no gross motor disturbances or dysarthria (Hall et al., 1993; Thoonen et al., 1996). These data were

collected using standardized tests of hearing level and language comprehension (Bomers & Mugge, 1989; Dutch translation of the Reynell Developmental Language Scales, Reynell & Huntley, 1985). From the 70 children referred by speech pathologists, only 19 children satisfied the above criteria completely and thus were considered ‘clear’ cases of DAS.³

In order to objectify the children who were ultimately selected for the study, language and speech characteristics of these children were extracted from the administered speech tasks (see the methods of Thoonen et al., 1996) and are shown in table 1.

Table 1. Individual scores on the selection tasks (meaningful and nonsense word-imitation task and a Maximum Repetition Rate task) in children with DAS and normally speaking children (NS).

DAS	Age	SQ	WQ	CQ	MemQ	Meaningful		Nonsense		MRR	
						Subst	Substpl	Subst	Substpl	Mono-syll	Tri-syll
#1	5;0	87	86	102	96	26%	76%	47%	68%	3.53	1.93
#2	5;1	85	88	92	89	24%	56%	64%	79%	4.12	3.38
#14	5;7	75	85	83	96	15%	60%	15%	30%	4.63	3.68
#17	5;10	72	80	83	82	17%	64%	67%	70%	4.63	Unable
#20	5;11	69	89	92	75	30%	60%	48%	72%	3.31	Unable
#21	5;11	73	94	89	96	31%	85%	-	-	3.20	Unable
NS	Age	SQ	WQ	CQ	MemQ	Subst	Substpl	Subst	Substpl	Mono-syll	Tri-syll
#54	4;9	-	-	-	-	7%	0%	12%	38%	4.29	3.52
#36	5;0	-	-	-	-	0%	0%	17%	64%	4.85	3.68
#42	5;3	-	-	-	-	3%	0%	6%	25%	4.74	4.05
#53	5;6	-	-	-	-	8%	0%	8%	20%	4.49	5.03
#58	5;6	-	-	-	-	5%	33%	9%	33%	4.44	5.08
#49	5;11	-	-	-	-	1%	0%	13%	0%	4.97	3.06

Note: SQ=sentence production quotient; WQ=word production quotient; CQ=comprehension quotient (>80 is normal); Mem.Q=Auditory Memory Quotient; Subst: percentage substitution of singleton consonants in syllable-initial position. Substpl: percentage substitution of place relative to the number of substitutions (Subst). Mono-syll: mean number of syllables per second (Maximum Repetition Rate, MRR) of monosyllable utterances (/pa/, /ta/, /ka/). Tri-syll: MRR in a tri-syllable utterance /pataka/.

The first two columns show the language production quotients at sentence (SQ) and word (WQ) level. The following columns display the language comprehension quotient and the auditory memory quotient (the average score of the normal population on these four quotients is 100; one standard deviation is 20). The columns entitled Subst and Substpl display the percentage substitutions of single, syllable initial consonants produced in an imitation task of meaningful and nonsense utterances. The percentage consonant substitutions are indicated with ‘Subst’; ‘Substpl’ indicates the percentage substitution of place-of-articulation relative to the total number of consonant substitutions (for example, in the word /pak/, the first phoneme is once substituted by /m/ resulting in /mak/ and once by /t/ resulting in /tak/. The latter is a substitution of

³ Obviously, all children who were referred by the speech-language pathologists showed apraxic characteristics, however, only those who satisfied all criteria were included in the present study.

place-of-articulation, which in this case equals 50% of the total number of substitutions). The Maximum Repetition Rates (MRR), expressed in mean number of syllables per second, are given in the last two columns (monosyllabic: /papa../, /tata../, /kaka../ and tri-syllabic: /pataka../).

Speech material

The speech samples consisted of disyllabic utterances of the type ‘/z/-V₁-/s/-/x/-V₂-/t/’, spoken within the carrier phrase ‘hé ... weer’ (/he-...-wI:r/) ‘Hey ... again’. In these utterances, V₁ was the vowel /ə/ in the open syllable /zə/ or the vowel /ʌ/ in the closed syllable /zʌs/, and V₂ was the vowel /a/, /i/, or /o/. We varied the syllable structure (syllable boundary is indicated with the symbol #) in these disyllabic utterances, in which the syllable boundary was either located before the /sx/-cluster or between /s/ and /x/, see table 2.

Table 2. List of the stimuli items (with translations), together with the syllable frequency of first and second syllable (based on the Dutch database CELEX consisting of 42 million lexical wordforms).

Syll. Struct.	Meaningful Utterance	Syll. Frequency		Nonsense Utterance	Syll. Frequency	
		First	Second		First	Second
#x	- zus giet – (‘sister pours’)	1413	394	- fus giek -	13	8
	- zus goot – (‘sister poured’)	1413	427	- fus gook -	13	0
	- zus gaat – (‘sister goes’)	1413	30067	- fus gaak -	13	0
#sx	- ze schiet – (‘she shoots’)	392675	1946	- de schiek -	3471753	0
	- ze schoot – (‘she shot’)	392675	3474	- de schook -	3471753	0
	- ze schaat-sen – (‘they skate’)	392675	143	- de scha-tel -	3471753	11343

Furthermore, syllable frequency was manipulated by using low(zero)-frequent syllables in nonsense utterances as well as high-frequent syllables in meaningful utterances. The manipulation of meaningfulness had a major effect on syllable frequency (see table 2).⁴ The low-frequent syllables were all possible Dutch syllables, in that they obeyed the phonotactic constraints of the language. Note that there is a slight articulatory difference between the vowels /ə/ and /ʌ/ (the latter can be articulated with more lip rounding than the first; International Phonetic Association, 1993). However, as will appear below, the value of the second formant frequencies of both vowels were equal.

⁴ The term ‘syllable frequency’ indicates the frequency of occurrence. In this study an extreme frequency manipulation was applied from nonsense utterances with low-frequent syllables and meaningful utterances with high-frequent syllables (CELEX, Baayen, Piepenbroek and Gulikers, 1995). This does not imply that all nonsense utterances consist of low-frequent syllables.

Procedure

The child repeated the utterances that were read aloud by the experimenter (the researcher who recorded all children). Since the focus of the study is on correct production and not on correct auditory perception, the experimenter used pictures of the utterances to support the production, in order to prevent articulatory incorrect production due to incorrect auditory perception. For example, a picture of a girl that was called ‘zus’ (/zʊs/) ‘sister’, watering the flowers, to support the utterance (within the carrier phrase): ‘hé zus giet weer’ (‘hey sister pours again’). In case of the nonsense utterances the child was instructed with pictures showing in one condition, ‘s#x’, a strange looking creature called ‘fus’ (/fʊs/) that said all kinds of funny things, like ‘giek’ (/xik/), ‘gook’ (/xok/), and ‘gaak’ (/xak/). In the other nonsense condition, ‘#sx’, the pictures displayed three non-existing animal-like creatures, which were called ‘schiek’ (/sxik/), ‘schook’ (/sxok/), and ‘schatel’ (/sxatəl/) respectively. These ‘nouns’ were preceded by the definite article ‘de’ (/də/) (‘the’).⁵

The utterances were elicited in blocks, each containing the three meaningful and three nonsense utterances within one syllable structure condition (‘s#x’ or ‘#sx’). If elicitation was completely randomized, the recording procedure would have been too confusing for the children, resulting in more erroneous productions. Between two blocks a cartoon was shown to the child as distraction and the experimenter and the child talked a little while about it. Within each session all utterance-types were repeated six times randomly, so in the end each child produced 72 utterances (3 vowels x 2 syllable structure conditions x 2 syllable frequency conditions x 6 repetitions). Sometimes children (especially the children with DAS) were not able to correctly produce an utterance. In that case (judged immediately by the experimenter) a second attempt was made immediately. This second (correct) production was, then, used for the acoustic analysis. Before the actual recordings were made each utterance type was practiced in order to prevent an additional learning effect during recordings.

A unidirectional dynamic microphone mounted on a headset (Shure SM10A) and a tape-recorder (Kenwood KX54) were used to record the speech samples. The headset kept the microphone at a constant distance of 5 centimetres in front of the right corner of the subject’s mouth. Recordings were made in a silent, though not soundproof, room at the schools of the children.

Acoustic analyses

The speech samples were digitized with a sample frequency of 25 kHz and the relevant sections, [V₁-/sx/-V₂] were spliced out, using the Kay Elemetrics Computerized Speech Lab (CSL) analysis system, Model 4300B. The speech samples were transcribed using a broad symbolic system (International Phonetic Alphabet), which was demonstrated to be a reliable method (Thoonen et al., 1994). Only the utterances in which [V₁-/sx/-V₂] was phonemically correct were used in the acoustic analyses. The percentage omission and substitution errors of the incorrectly produced utterances will be displayed in the results section.

⁵ It should be noted that the syllable ‘de’ (‘the’) is high frequent. However, the total utterance is nonsense.

As a first step in the acoustic analysis, the onset and offset of each segment were marked by inspection of the oscillogram, the FFT (Fast Fourier Transformation) spectrogram, and the energy-window. In the oscillogram, the CSL-program automatically produced impulse markers in the voiced sections (V_1 and V_2) of the signal at the zero-crossings in the steep rises of the curve, corresponding with the closing of the glottis during voicing (wrongly placed impulse markers were corrected interactively). The onset and offset of the vowels were determined by using these voicing impulse markers and the information in the spectrogram (for presence of a formant structure). The offset of voicing (absence of fundamental frequency and formant structure) and the beginning of a noise structure in the spectrogram determined the end of V_1 , which corresponded with the beginning of [s]. To determine the transition from [s] to [x] information of the spectrogram was used visually along with the energy-contour. The fricatives can be distinguished by their energy distribution in the spectrogram, that is the [s] contains most energy in the high frequency regions, whereas in the consonant [x] more energy is found in the lower frequency regions. As a consequence, the overall energy, displayed in the energy-contour, is higher in [s] than in [x]. With this information of the spectrogram and energy-contour, the location of the transition from [s] to [x] could be determined by hand. Figure 1 displays an example of an oscillogram with the on- and offset markers (see triangles), the energy contour, and the FFT-spectrogram with first and second formant traces.

Inter-observer agreement concerning the placing of these markers by two observers was tested in a subset of the data. Each observer placed markers in the utterances of three NS children and three children with DAS. The average difference between the markers set by the different observers was not larger than 7.7 ms. High correlation coefficients were found between the observers (from 0.97 to 0.99 for the different marker-placements), which indicated a significant reliability between observers, and of the method used to place the markers.

F2 values were extracted at seven locations in the signal: at V_1 midpoint (1), at the end of V_1 (2), in the consonant [s] (3), in the consonant [x] (4), at the beginning of V_2 (5), at the end of the transition in V_2 (6) and at V_2 midpoint (7). In the voiced sections, V_1 and V_2 , the formant values (with maximum bandwidth of 600 Hz) were obtained using pitch-synchronous Linear Predictive Coding (LPC) analyses (triangular analysis window; 20 components autocorrelation with pre-emphasis of 0.950), followed by a root-solving procedure (Nijland et al., 2002). Additional to the F2-measurements in the vowels, two separate LPC-analyses were performed close to the offset of the unvoiced segments [s] and [x]. For this, F2 was calculated from a window (20 ms width) centred 20 ms before offset in the consonants [s] and [x]. Besides extracting the F2 values from the signal, also the durations of all segments, V_1 , /s/, /x/, V_2 , were determined using the markers at the beginning and end of each segment.

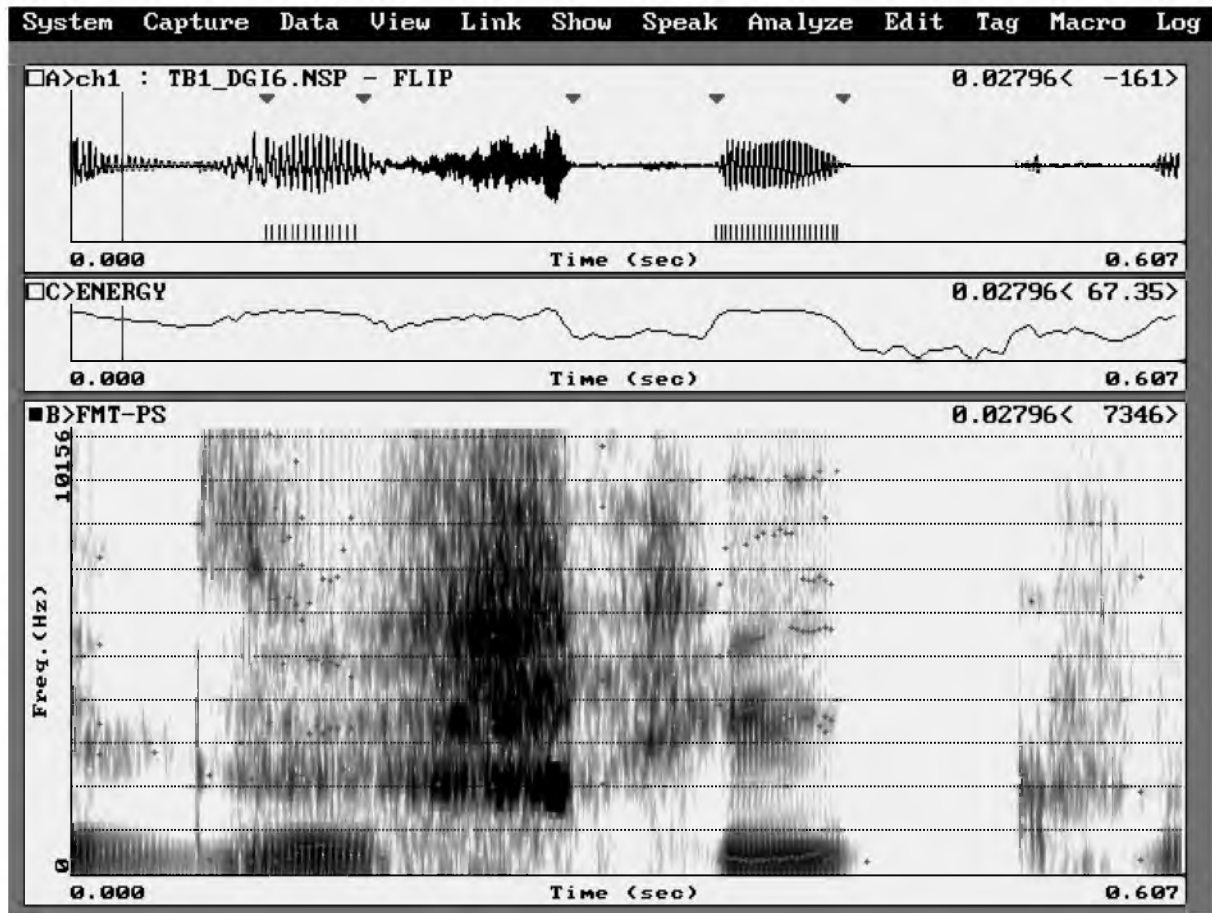


Figure 1. Oscillogram (uppermost window) and spectrogram (lowermost window) of the utterance /he: dæ#sɪkwI:r/. The little triangles in the oscillogram display the markers that were placed at segment transitions; the small black dots in the spectrogram represent the formant values.

Anatomical differences between speakers are expected to result in large inter-speaker variances in mean formant values across vowels. To correct for this variability, as a means of speaker normalization, formant *ratios* per utterance type were calculated for each child separately. For each syllable structure, the formant values (of six repetitions) of utterances with /i/ were averaged and divided by the average formant values of utterances with /o/ (i/o-ratio).⁶ Subsequently, these i/o-ratios are used to determine the coarticulation effect of the vowel on preceding phonemes, as an index for distinction between the utterances: the higher the ratio, the larger the distinction. Typically, large ratios are found at vowel midpoint, where the difference between the utterances is at maximum. Smaller ratios close to unity, such as typically found in the schwa, reflect that F2 values are equal, independent of the upcoming vowel (no coarticulation). Thus, the higher the ratio is above 1 the larger the distinction is between the utterances at that particular location.

⁶ Also i/a-ratios were calculated. Overall, the value of the i/a-ratios was lower than of the i/o-ratios, as expected, but their patterns were similar. The i/a-ratios are not presented in the figures.

Statistical design

In order to answer the question whether syllable structure and syllable frequency influence coarticulatory cohesion and durational pattern, analyses of variance are conducted on the i/o-ratios and the segment durations in both groups of speakers. In these analyses of variance the between subject factor is 'Group', and the within subject factors are 'Syllable structure' and 'Syllable frequency' (including all interactions).

Apart from the interaction effect of syllable structure or syllable frequency with coarticulation, also the main effects of syllable structure and of syllable frequency on the (raw) F2-values in both groups are evaluated as well as the main effect of coarticulation of the second vowel on preceding segments. Before performing analyses of variance the condition of homogeneity of variance in the groups that are to be compared must be satisfied. This was a problem in the formant frequency data and the durational data: the repetitions in the children with DAS contained more variance as compared to the NS children (which will be discussed in the results and discussion section). Furthermore, children with DAS were not always able to correctly produce six repetitions of each utterance-type. In order to statistically correct for this problem of heterogeneity we aggregated the repeated utterances per child, which resulted in average data of each utterance type per child. On these aggregated F2 data analyses of variance were conducted, with the factors 'Group' (between subject factor), 'Syllable structure', 'Syllable frequency', 'Vowel2' (within subject factors). Furthermore, in order to evaluate the main effect of coarticulation of the second vowel on preceding segments, a one-sample *t*-test is conducted to test whether the i/o-ratios were significantly higher than 1.

RESULTS AND DISCUSSION

The effect of the manipulation of syllable structure and syllable frequency in children with DAS when compared to NS children will be presented and discussed on (1) phonemic error frequencies, (2) coarticulation patterns in formant measures (F2), and (3) relative segment durations. Furthermore, global differences, concerning variability, between children with DAS and NS children will be discussed at the end of this section.

Syllable structure and syllable frequency

Phonemic errors

The utterances were broadly transcribed, which showed that whereas NS children did not exhibit problems (error percentages close to 0), some children with DAS showed high error frequencies. Errors appeared particularly in the /sx/-sequence (in both 's#x' and '#sx'), even though in a different speech task, recorded during the same session, the children demonstrated that the singleton /s/ and /x/ were in the children's repertoire (Nijland et al., 2002). Table 3 shows for each syllable structure ('s#x' and '#sx') the percentage of correct utterances produced by the children with DAS and the percentage of phonemic errors on /s/ and /x/. The error percentage of the high-frequent and low-frequent utterances were aggregated in the table (the patterns were not different; on Wilks' Lambda based $F(2,43) = 0.070$, n.s.). The column 'omission' contains the

percentage ‘s#x’-utterances in which either /s/ or /x/ is omitted; the column ‘cluster reduction’ contains the percentage ‘#sx’-utterances in which the cluster was reduced to either [s] or [x]. The pause is defined by the absence of energy in the signal (i.e., absence of speech), during at least 8 ms.⁷

Table 3. Percentages errors in the sequence /s-x/ produced by children with DAS. The low percentage of ‘other errors’, like vowel errors, is not displayed in this table

Target production	s#x			#sx		
Production	Correct	Omission	Pause	Correct	Cluster reduction	Pause
Child 1 (5;0)	25.0	41.7	33.3	5.6	83.3	11.1
Child 2 (5;1)	94.4	2.8	0	0	97.2	0
Child 14 (5;7)	97.2	0	2.8	91.6	0	5.6
Child 17 (5;10)	97.2	0	2.8	97.2	2.8	0
Child 20 (5;11)	66.7	16.7	11.1	47.2	47.2	5.6
Child 21 (5;11)	52.8	33.3	11.1	5.6	88.9	0

Table 3 shows that some children exhibit a very low percentage of correct productions (children no. 1, 2, 21). Remarkable are the children that were able to produce the sounds /s/ and /x/ successively, without pause, in the condition with the syllable boundary in between the two sounds (‘s#x’), yet not in the condition with the syllable boundary preceding the cluster (‘#sx’). Child 2 distinctly shows this effect: 94.4% correct ‘s#x’-productions, but with no correct productions of ‘#sx’. A similar difference was found in the children 1, 20 and 21, although to a lesser extent. Thus, four out of six children with DAS showed an effect of syllable structure on phonemic /sx/ production. Apparently also in children with DAS the speech production is characterized by a syllabic organization.

Furthermore, table 3 shows that a pause between /s/ and /x/ occurred more often in the condition ‘s#x’ than in ‘#sx’. Normally, production of a pause between words or syllables (in the sequence /s#x/) is possible without affecting the syllable structure of the utterance. However, if a pause is produced between /s/ and /x/ in the /#sx/-utterances, this will affect the syllable structure and consequently the meaning of the utterances, in that ‘#sx’ changes to ‘s#x’. Three children with DAS (children 1, 14, 20) produced substantial percentages of pauses within the cluster [sx] (/#sx/). In contrast, none of the NS children produced pauses within the cluster. In terms of speech production processes these pauses within the cluster, contradictory to syllable structure effects above, could be interpreted as an indication of segmental rather than syllabic planning.

⁷ In normal speech the fricatives unit seamlessly: a silence of 8 ms provides sufficient information of a pause.

Formant measures

The high percentages of production errors in children 1, 2, 21 induce a problem to further process the acoustic data of these children, because some utterance types are completely missing (e.g., the ‘#sx’-utterances in child 2). The average F2-trajectories of the remaining (three) children with DAS and of the NS children are displayed in figure 2; utterances with high-frequent syllables at the left-hand side, low-frequent syllables at the right-hand side; the solid lines represent the ‘s#x’-utterances, the dotted lines the ‘#sx’-utterances.

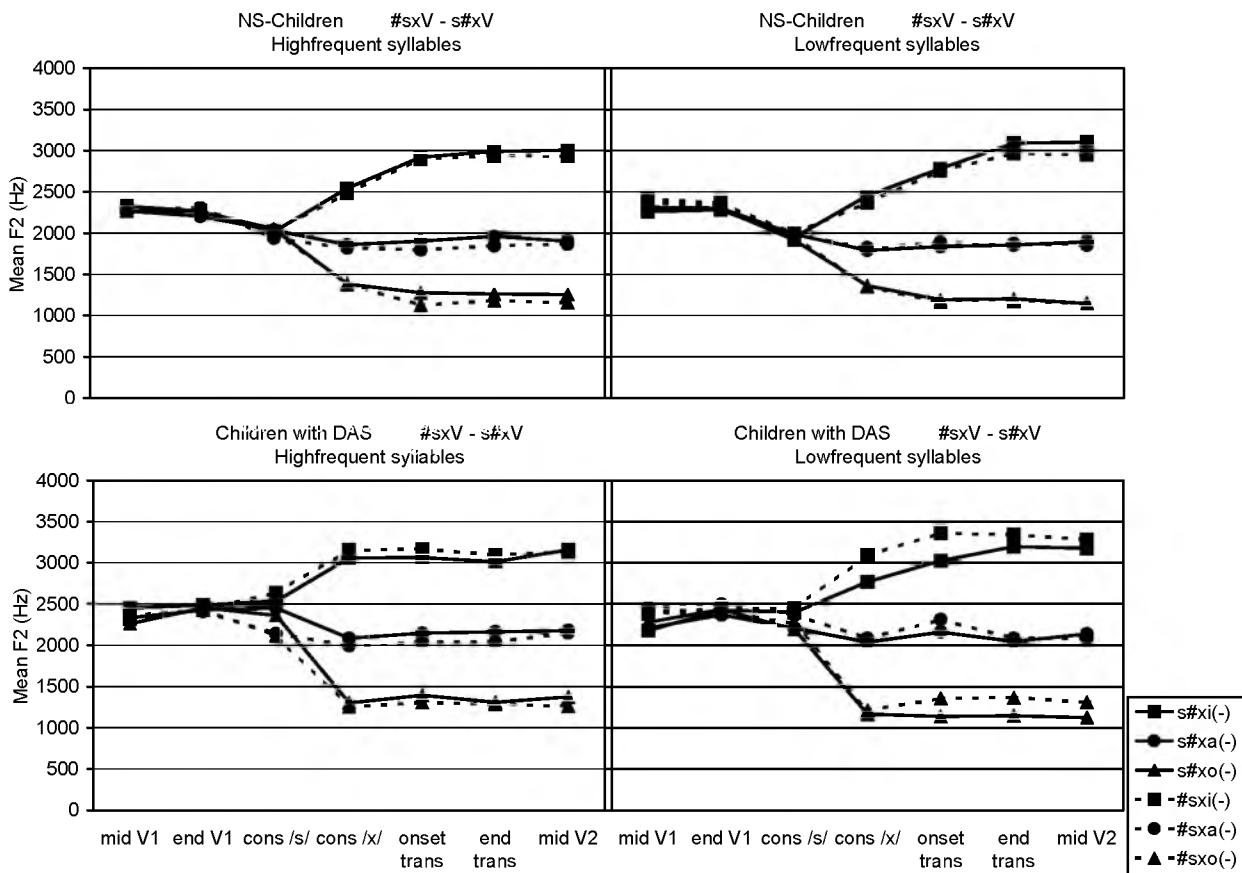
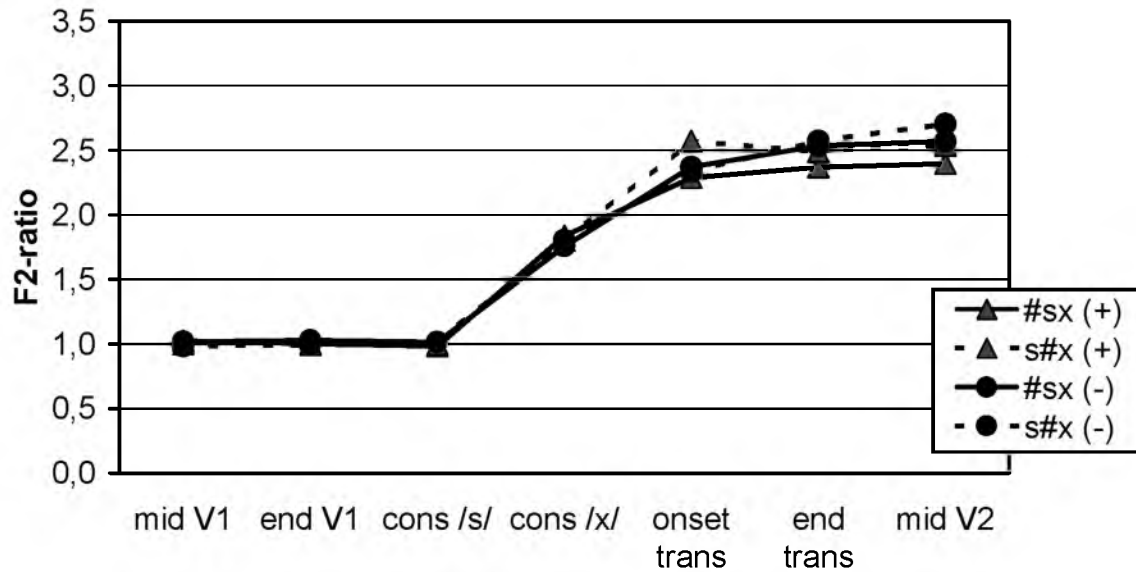


Figure 2. F2-trajectory of high-frequent syllable (left-hand side) and low-frequent syllable utterances (right-hand side) produced by normally speaking children (upper figures) and children with DAS (lower figures).

Before discussing the interaction effect of syllable structure on coarticulation, we go through the main effect of syllable structure and the main effect of coarticulation. An effect of syllable structure is visible in figure 2 (dotted versus solid lines), especially in children with DAS. Figure 2 shows that F2 values were higher in ‘#sx’ than in ‘s#x’ at mid V1 ($F(1,7) = 7.04, p < 0.05$), at transition onset ($F(1,7) = 5.87, p < 0.05$), and at mid V2, ($F(1,7) = 7.32, p < 0.05$); the latter effect of syllable structure is stronger in children with DAS than in NS children, $F(1,7) = 6.31, p < 0.05$. It is hard to explain why higher F2-values are found in ‘#sx’ versus ‘s#x’ (and especially in children with DAS). Figure 2, furthermore, shows that, as expected, differences between the F2-

frequencies, due to differences in second vowel, are large in V_2 , indicating a large differentiation of the vowels [a], [i], and [o], and smaller in the preceding segments, representing the effect of anticipatory coarticulation of the upcoming vowel V_2 . As a means of speaker normalization, and as an index of coarticulation, i/o-ratios are calculated.

NS-Children i/o-ratios



Children with DAS i/o-ratios

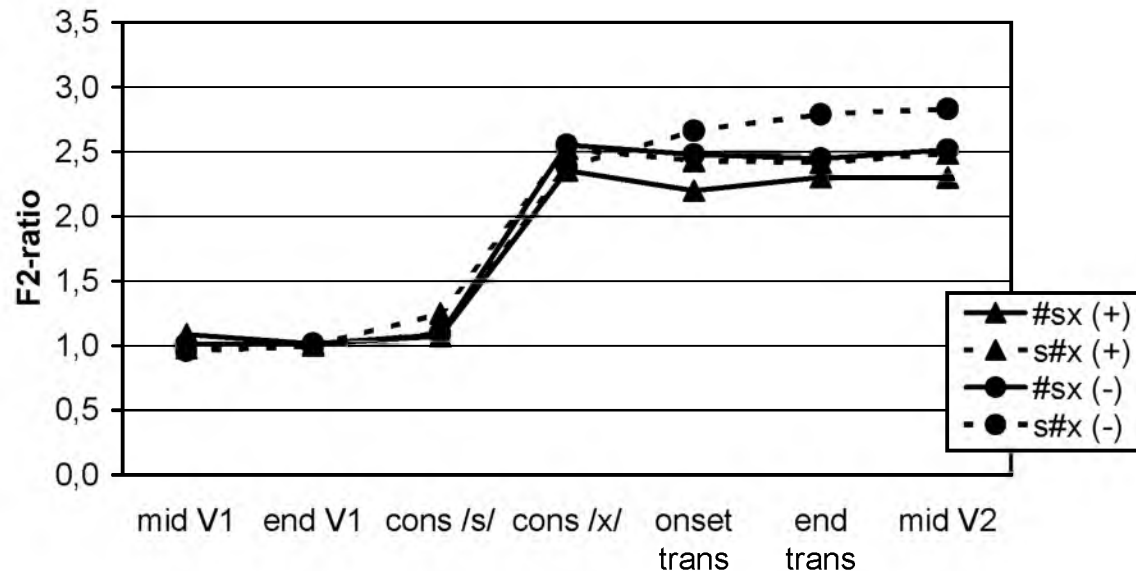


Figure 3. i/o-ratio's in normally speaking children (upper figure Figure 3a) and children with DAS (lower figure Figure 3b) in high-frequent syllable (+) as well as low frequent syllable (-) utterances.

Figure 3 shows the i/o-ratios in the successive locations of the NS children (3a) and the children with DAS (3b). The ratio-figures of both groups show a very small coarticulation effect from V_2 on V_1 (ratio is close to 1) and a larger coarticulation effect from V_2 on the [x]. Also, right at the onset of V_2 , the i/o-ratio is high. In NS children (figure 3a) the coarticulation of the vowel V_2 was significant from [x] onward (i/o-ratios higher than 1: all $t(22) > 11.56$; $p < 0.001$). In children with DAS (figure 3b) ratios significantly higher than 1 were found from [s] onward (all $t(11) > 3.17$; $p < 0.01$). Moreover, the result of an analysis of variance showed higher ratios in consonant [x] in children with DAS as compared to NS children (Group: $F(1,7) = 11.77$; $p < 0.025$), which indicates that intra-syllabic coarticulation was stronger in children with DAS. The i/o-ratios in the middle of V_2 , which reflect the differentiation made between the vowels, did not differ between children with DAS and NS children ($F(1,7) = 0.08$; n.s.).

Thus, the significant coarticulation effect from the vowel V_2 on the preceding segments is earlier (significant from [s] onward) and stronger in children with DAS as compared to NS children. This is an indication for stronger intra-syllabic coarticulation in DAS as compared to normal speech. Nittrouer et al. (1996) showed that during normal speech development the anticipatory coarticulation of a vowel on preceding sounds decreases with increasing age. From a gestural interpretation this means that the overlap between gestures in young children is larger than in older children and adults. According to this view, the results of the children with DAS in the present study (stronger anticipatory coarticulation and earlier in the utterance) might be interpreted as an indication for delayed development.

The main question in the present study is whether differences in anticipatory (intra- and inter-syllabic) coarticulation appear as a consequence of manipulating the syllable structure in NS children and, more interestingly, in children with DAS. In the figure 3, such an effect is primarily expected in the consonant that changes syllable position, that is in the consonant [s]. Stronger coarticulation is expected if the /s/ belongs to the same syllable as the vowel V_2 (in ‘#sx’-utterances), than if it is part of the preceding syllable (in ‘s#x’-utterances).

The i/o-ratios (figure 3) show a significant effect of syllable structure on coarticulation at the location mid V_1 ($F(1,7) = 6.11$; $p < 0.05$), showing that *inter*-syllabic coarticulation in ‘#sx’ is stronger than in ‘s#x’. This effect emerges in both children with DAS and NS children. Otherwise, syllable structure does not affect (inter- and intra-syllabic) coarticulation at other measurement points (all $F(1,7) < 1.09$, n.s.). In explaining the effect of syllable structure on V_1 , the quality of V_1 seems to be important. Although the average second formant values of V_1 in the open syllable context (/zə/ and /də/), are equal to the values in the closed syllable (/zʌs/ and /fʌs/), there seem to be other differences. The syllables differ in phonological specification of V_1 and in prosodic sense, which might influence the inter-syllabic coarticulation in two ways. First, the phonologically more specified vowel /ʌ/ possibly allows for less coarticulation than the neutral vowel /ə/. This finding corresponds to the principle of underspecification (Keating, 1988), which suggests that phonologically unmarked features remain unspecified in phonetic realizations. This enables forward and backward feature spreading of surrounding phonemes onto preceding or following underspecified phonemes. For example, since the feature ‘lip rounding’ is not specified (or marked) in the [s], [s] is vulnerable to anticipatory lip rounding of a

following [u]. Thus, the underspecified /ə/ is easily influenced by context (the following/preceding consonant and vowel), as opposed to /ʌ/.

Second, besides the difference in phonological specification between the vowels, the syllables differ in prosodic sense as well. The significant difference in the i/o-ratios, between spondaic (as in 'CV₁s#xV₂C') and iambic utterances (as in 'CV₁#sxV₂C') found at mid V₁, corroborates results of other studies in which vowels in prosodically stronger positions exhibited less coarticulation than in weaker positions (Cho, 1999; De Jong, Beckman, & Edwards, 1993). The closed syllables (/CV₁s#/) are stressed, whereas the open syllables (/CV₁#s/) do not have stress in these utterances. Both interpretations, phonological underspecification as well as difference in prosody processing, might account for the effect of syllable structure on the coarticulation in V₁.

To sum up, changing the syllable structure did not affect the strength and the extent of *intra*-syllabic coarticulation from the upcoming vowel V₂ in the crucial phoneme /s/, neither in NS children nor in children with DAS. In contrast, a significant effect of syllable structure on coarticulation was found on formant ratios at mid V₁ in both groups, which indicates stronger inter-syllabic coarticulation in '#sx' than in 's#x'.

The second question in this study was whether syllable frequency influences the coarticulatory cohesion in the utterances. Under the assumption that the articulatory gestures of frequently used syllables are stored in the syllabary and therefore will have more cohesion, we expect to find stronger coarticulation in high-frequent syllable utterances when compared to low-frequent syllables. Before discussing the interaction effect of syllable frequency on coarticulation, we briefly mention the main effect of syllable frequency on F2 values. Figure 2 shows that manipulation of syllable frequency results in significantly higher F2 values at consonant [s] in the high-frequent utterances as compared to low-frequent utterances ($F(1,7)=21.0$; $p<0.005$). However, this syllable frequency effect was independent of the upcoming vowel V₂.

An interaction of syllable frequency with coarticulation is not found. Syllable frequency does not affect the strength or extent of the coarticulation (see figure 3), neither in NS children nor in children with DAS (all F s (1,7) < 1.11; n.s.). From the absence of a syllable frequency effect in both groups we must conclude that the existence of a syllabary, including a possible deficient syllabary in children with DAS, cannot be substantiated. This could mean that either the assumption of a syllabary is false or that the effect of the syllable frequency manipulation was not strong enough in our data. Note that in the construction of the speech material only the frequency of the second syllable was strongly manipulated, especially for the syllables used in the i/o-ratios.

Durational measures

The speech of children with DAS is characterized by slow speaking rate, that is, long segment durations. Figure 4 displays the average segment durations of the different utterances types (figure 4a, NS children; figure 4b children with DAS). The results of the analysis of variance on the durations of the total group and the two groups separately (NS children and children with DAS) are displayed in table 4. Both the figures and the results of the analysis of variance clearly show that children with DAS have significantly longer total and segment durations as compared to NS children (except for V₂).

Furthermore, an effect of syllable structure emerges in the durations. This effect is different in children with DAS as compared to NS children. In NS children (figure 4a) the '#sx'-utterances are significantly shorter than 's#x'-utterances. The reason for the shorter total duration of the '#sx'-utterances is due to a shorter V_1 , [x], and V_2 (shorter in '#sx' than in 's#x'). In the durations of children with DAS (figure 4b) a significant effect of syllable structure is only found in the segment duration of V_2 (shorter in '#sx' than in 's#x'), but this does not result in significantly shorter total durations.

Table 4. *F-values on segment durations across groups and of the two groups separately. Presented are F-values from the multivariate analysis of variance with main effects and interaction effects of the factors group, syllable structure (Syl.struct.), vowel2, syllable frequency (Syl.Freq.). The direction of the significant effects is displayed between square brackets*

Group	Factor	df ₁ , df ₂	Total dur.		V_1		/s/		/x/		V_2	
			F	Sign.	F	Sign.	F	Sign.	F	Sign.	F	Sign.
Total	Group (G)	1,7	16.55**	[NS<DAS]	9.79*	[NS<DAS]	8.52**	[NS<DAS]	14.62**	[NS<DAS]	4.36	
	Syl.struct. (SS)	1,7	15.31**	[#sx<s#x]	14.45**	[#sx<s#x]	1.95		14.82**	[#sx<s#x]	13.36**	[s#x<#sx]
	Vowel2	2,14	4.96*	[/i/</a,o/]	0.68		64.22**	[/a,o/</i/]	5.16*	[/a,o/</i/]	92.15**	[/i/</a,o/]
	Syl.Freq. (SF)	1,7	2.11		4.44		4.60		0.89		4.75	
	G * V2	2,14	2.86		0.32		5.19** ²⁾		4.05* ²⁾		1.54	
	G * SF	1,7	1.21		3.58		0.98		1.08		1.74* ⁴⁾	
	G * SS	1,7	1.58		17.05** ¹⁾		6.73** ³⁾		0.71		0.33	
DAS	Syl.struct. (SS)	1,2	0.95		0.28		9.74		4.13		346.44**	[#sx<s#x]
	Vowel2	2,4	0.04		0.80		19.58*	[/a,o/</i/]	2.56		26.97**	[/i/</a,o/]
	Syl.Freq. (SF)	1,2	5.10		51.19*	[high<low]	1.40		1.06		4.50	
NS	Syl.struct. (SS)	1,5	30.93**	[#sx<s#x]	45.74**	[#sx<s#x]	0.83		10.29*	[#sx<s#x]	7.35*	[#sx<s#x]
	Vowel2	2,10	18.33**	[/i/</a,o/]	0.06		57.56**	[/a,o/</i/]	0.65		66.86**	[/i/</a,o/]
	Syl.Freq. (SF)	1,5	0.10		0.06		3.15		0.00		4.13	

Note. 1) The effect of syllable structure is stronger in NS children than in children with DAS (ns. in DAS).

2) The effect of vowel2 is stronger in children with DAS than in NS children.

3) The effect of syllable structure (s#x<#sx) is stronger in children with DAS than in NS children (ns. in NS).

4) The effect of syllable frequency (highfrequent<zero frequent) is stronger in children with DAS than in NS children (ns. in NS). $p < 0.05$; ** $p < 0.01$

Thus, the NS children show strong, significant effects of syllable structure on the durations of three of the four segments. Two different effects seem to be operative here: a metric and a prosodic effect. First, with regard to the duration of the consonant [x], it is found that the consonant [x] is shorter in the cluster '#sx' than in syllable-initial position in '#sx'. It is suggested that the duration of the consonant [x] is adjusted to the change in metrical structure of the second syllable. This means that in the syllable /#sx V_2 / the duration of [x] is shorter than in the syllable with /#x V_2 /, because of the extra segment /s/ in /#sx V_2 /. The duration of V_2 is similarly adjusted to this change in metrical structure (shorter in '#sx' than in 's#x'). Second, the duration of vowel V_1 (in the first syllable) depends on syllable structure: V_1 is shorter in /CV₁#sx V_2 C/ than in /CV₁s#x V_2 C/. This effect cannot be explained by a change in metrical

structure. Yet, in these utterances the first syllables differed in prosodic sense dependent on the syllable structure. That is, the closed syllable ($/CV_1s\#/$) is stressed, whereas the open syllable ($/CV_1\#/$) does not have stress. Thus, the significant difference in duration between spondaic (as in ' $CV_1s\#xV_2C$ ') and iambic utterances (as in ' $CV_1\#sxV_2C$ ') found in V_1 for the NS children corresponds with shorter durations of vowels in prosodically weaker positions.

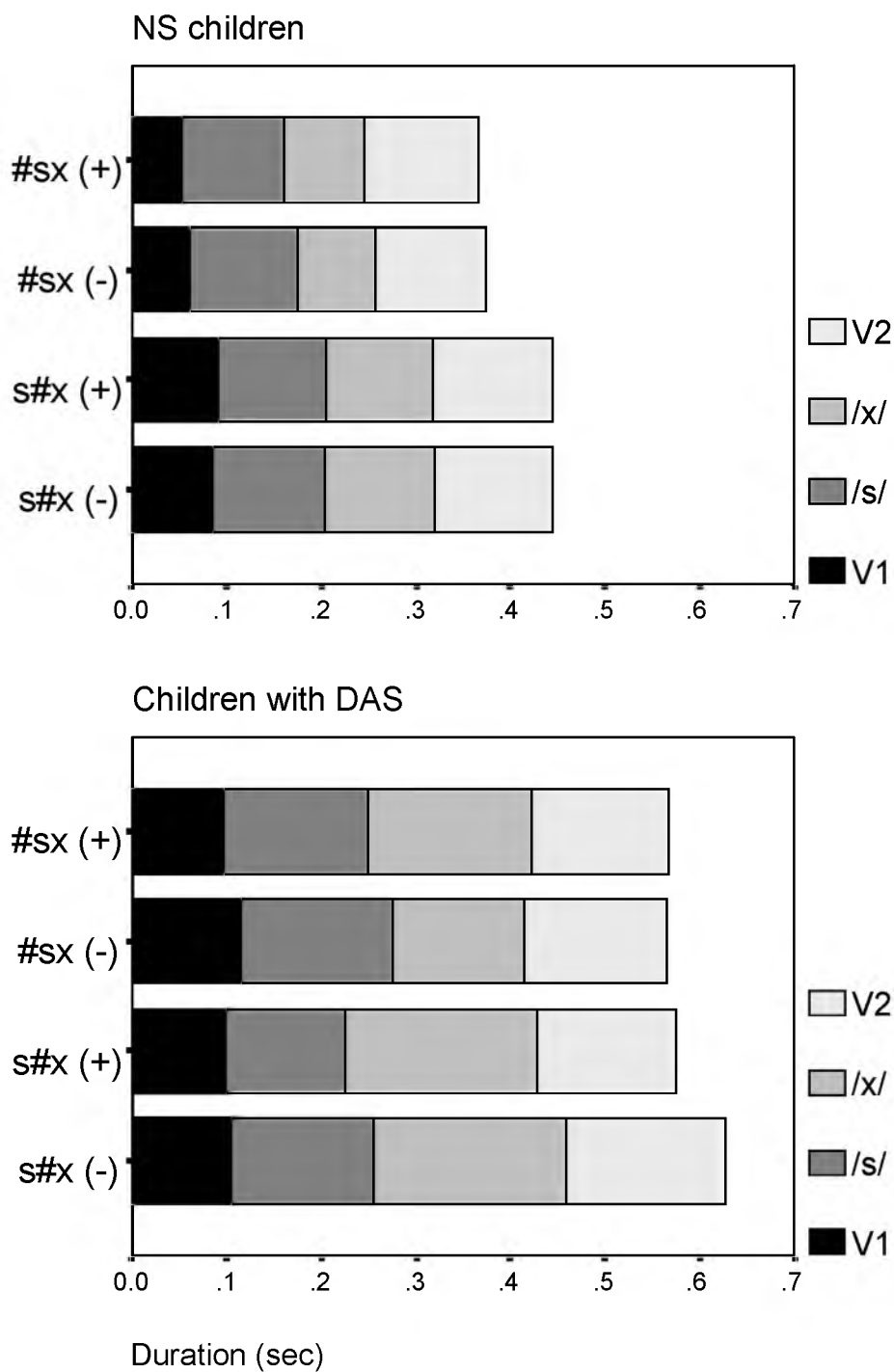


Figure 4. Mean segment duration of normally speaking children (upper figure Figure 4a) and children with DAS (lower figure Figure 4b).

Although, as compared to the NS children, the children with DAS show a similar effect of syllable structure on the duration of V_2 , no significant effect of syllable structure is found in [x]. Thus, in children with DAS adjustment to the change in metrical structure of the second syllable is only effectuated in V_2 . An effect of syllable structure on the duration V_1 , as was found in NS children, is not found in children with DAS. We argue that this difference between NS children and children with DAS is a difference in the way these two groups process prosodic aspects. NS children shorten the duration of V_1 in the prosodically weaker position. The fact that children with DAS do not show a significant effect of syllable structure in V_1 (the effect of syllable structure on the duration of V_1 even tends to go in the opposite direction) is an indication for deviant prosodic patterns in children with DAS. This confirms the findings of Shriberg et al. (1997) and Velleman and Shriberg (1999), who reported inappropriate sentential stress in children with DAS. They showed that children with DAS made less distinction between stressed and unstressed segments as compared to NS children. An explanation for this could lie in difference in suprasegmental processing of the phonological phrase.

Syllable frequency does not account for differences in durational structure, except for a longer duration of V_1 in children with DAS in the low-frequent syllable utterances as compared to the high-frequent syllable utterances. As was mentioned above, syllable frequency also did not influence coarticulation as measured in the i/o-ratios. The absence of a strong effect of syllable frequency either in durational structure or in coarticulation, together with the above-mentioned comment on the manipulation of syllable frequencies, makes it hard to draw conclusions from the syllable frequency results. However, the suggestion that in children with DAS the processing of the phonological phrase is different compared to NS children continues to exist.

Besides the effect of syllable structure and syllable frequency on the durational pattern, table 4 shows that the total durations and segment durations differ significantly due to the factor Vowel2. A significant shorter duration of the vowel [i] leads to shorter total durations of the utterances with /i/, and to longer durations of the preceding segments in the /i/-utterances (except for V_1). In the separate group analyses, this effect of Vowel2 is found in the durations of the segments V_2 and [s] in both groups. However, in contrast to NS children, a shorter duration of the vowel [i] does not result in significantly shorter total durations of the utterances with /i/ in children with DAS.

Global differences: variability

Table 5 displays the variability in formant frequency values and durations measured in the two groups of speakers. The standard deviations are calculated over the average values per speaker (between speaker variability), over the repetitions within one speaker (within speaker variability), and over different utterance types. For the F2 values, this was done at each location (from mid V_1 to mid V_2), and for the durational measures the standard deviation was calculated for each segment (V_1 , [s], [x], V_2). The significance of difference in variability between the two groups is calculated with *F*-ratios on the variances (viz. the square of the standard deviation). These calculated *F*-ratios are displayed underneath the standard deviation in the table.

Table 5. Standard deviations calculated at each location, due to utterance-type (Type), speaker or between subject variability (Speaker), and within subject variability (Within). F-ratios of between and within subject variability are calculated to show differences between speaker groups.

Group	Factor	df	Standard Deviations of F2 values (Hz) at the locations						
			mid V ₁	end V ₁	Cons /s/	Cons /x/	begin trans	end trans	mid V ₂
DAS	Type	11	317.2	132.9	609.1	3091.0	3396.7	3543.4	3742.6
	Speaker	5	1223.0	1733.6	1358.6	557.8	735.2	841.6	780.7
	Within	210	220.2	186.6	274.7	305.4	284.9	247.9	219.4
NS	Type	11	313.1	242.6	176.6	2550.7	4140.5	4439.6	4713.1
	Speaker	5	1521.0	1196.6	1379.4	1201.2	1015.0	704.6	787.7
	Within	380	176.6	158.9	172.6	233.3	256.5	225.1	210.7
DAS/NS F(speaker)			1.55	2.10	1.03	4.64	1.91	1.43	1.02
DAS/NS F(within)			1.55 **	1.38 **	2.53 **	1.71 **	1.23 *	1.21	1.08
Group	Factor	Df	Standard Deviation of segment durations (ms)						
			V ₁	/s/	/x/	V ₂			
DAS	Type	11	43.6	83.5	107.3	203.2			
	Speaker	5	144.1	170.8	257.1	194.3			
	Within	230	26.1	43.2	53.8	33.6			
NS	Type	11	102.3	56.7	97.7	201.8			
	Speaker	5	114.4	114.4	135.6	176.7			
	Within	420	18.8	21.0	26.2	20.6			
DAS/NS F(speaker)			1.59	2.23	3.59	1.21			
DAS/NS F(within)			1.92 *	4.24 *	4.20 *	2.67 *			

Note: significance: * $p < 0.05$; ** $p < 0.01$.

The standard deviation of the F2 values show that the variability due to type of utterance increased to the end of the utterance in both groups, which is explained by the difference in utterance-types in V₂. Variability due to utterance type is not significantly different between the groups (see *F*-values). The standard deviation between speakers decrease from the beginning to the end of the utterance, although not monotonically. Again, the groups do not differ significantly on variances due to the factor Speaker, which means that the children with DAS do not differ more strongly among themselves than NS children do. In both groups the within speaker standard deviation (due to repeated productions) is highest in the middle of the utterances: in consonant [x] for the children with DAS and at the beginning of the transition in V₂ for the NS children. This within speaker variability is significantly larger in children with DAS as compared to NS children at all locations except for the steady state of V₂ ('transition end' and 'mid V₂'). This indicates that children with DAS produce acoustically more variable repetitions than NS children.

The standard deviation of the segment durations, displayed in the second half of table 5, show a similar pattern when compared to the standard deviations of the F2 values. However, the standard deviation between speakers show an increase to the end of the utterance (again not

monotonically) instead of a decrease. The children with DAS are again more variable in the segment durations of their repeated utterances as compared to NS children. Again, the variability between speakers is not larger in children with DAS than in NS children.

To summarize, children with DAS display high variability in acoustic measures of phonemically correct utterances, which appear in the variances of repeated productions. This inconsistency in repetitions can be interpreted as evidence of immature or disturbed speech motor control (Nijland et al., 2002). In general we can conclude that children with DAS exhibit problems in using the syllables in the automation process of speech production. These results not only contribute to the diagnosis and understanding of DAS, but also yield guidelines for therapy. The small speech elements focused on in training programmes for children with DAS are the size of syllables in order to enhance automation (the Dutch Dyspraxia Program, Erlings-van Deurse, Freriks, Goudt-Bakker, Van der Meulen, & de Vries, 1993); based on the Nuffield Dyspraxia Programme, Connery, 1992). The results of the present study support this approach, however, more research is needed to evaluate the effectiveness.

In contrast to the difference in within speaker variability between children with DAS and NS children, the variance between speakers is not significantly larger in the children with DAS than in the NS children. This result was not expected. However, we have to keep in mind that between speaker differences are tested on the variances of the isolated measurement points and not on the patterns. Thus, the often-reported heterogeneity within the group of children with DAS could not be corroborated by the variability results in the present study.

CONCLUSION

In the present study we investigated whether the deviant coarticulation patterns (as found in previous research, Nijland et al., 2002) reflect an impairment in the planning of syllables in DAS. In particular, the main question was whether children with DAS as compared to NS children show deviant patterns in intra-syllabic and inter-syllabic coarticulation and durational structure as a consequence of syllable structure (and syllable frequency). If so, this would indicate a disturbance in the planning of syllables, including a deficient use and/or accessibility of the syllabary (a repository of frequently used syllables). In order to find an answer to this question, the second formant (F2) trajectory and segment durations were measured in phonemically identical utterances with systematic differences in syllable structure ('CV₁#sxV₂' versus 'CV₁s#xV₂') and syllable frequency (highfrequent versus lowfrequent syllables).

The coarticulation patterns of both groups showed that coarticulation is stronger and more extended in children with DAS when compared to NS children. However, this effect is independent of syllable structure. We argued that this result of stronger and more extended coarticulation in children with DAS is an indication for delayed development.

An effect of syllable structure on coarticulation of V₂ on the preceding /s/ could not be substantiated, neither in NS children nor in children with DAS. Although a differential coarticulation effect due to syllable structure is found in the i/o-ratios of V₁ (the coarticulation effect of V₂ was stronger in the utterances with '#sx' than in 's#x'), no differences are found between the two groups. We argue that a difference in phonological specification of the first

vowel (/ʊ/ is more specified than /ə/, and therefore less vulnerable for coarticulation) or prosody processing (in which vowels in prosodically stronger positions exhibit less coarticulation) is underlying this effect of syllable structure on V_1 .

In the durational structure, more than in the coarticulatory patterns, effects of syllable structure and differences between NS children and children with DAS did emerge. In NS children the shortening of the [x] and V_2 in the '#sx'-utterances as compared to the 's#x'-utterances is interpreted as an adjustment to the metrical structure of the second syllable that contains an extra segment /s/ in the syllable /#sx V_2 / as compared to the syllable /#x V_2 /. In children with DAS a similar effect is found in V_2 , but not in /x/. In addition to the shorter duration of V_2 in '#sx' as compared to 's#x', the results on error percentages, which indicate that the production of [#sx] induced more difficulty than [s#x], support the hypothesis that also in children with DAS the speech production process passes through a level which is characterized by a syllabic organization. These syllabic effects in DAS were, however, not as systematic as in normal speech. Furthermore, in contrast to NS children, children with DAS show no effect of syllable structure on the duration of V_1 . This finding is furthermore discussed to be a problem in prosodic processing in DAS. Models on speech production suggest that information about metrical structure and prosody are combined in what is called the 'phonetic encoding' (Kent, 2000; Levelt et al., 1999), 'phonetic program assembly' (Ozanne, 1995), or 'motor planning' (Van der Merwe, 1997); to our opinion these are all different names to specify the same stage in which the motor plan of the syllable is generated. Thus conceived, the data of the present study revealed that children with DAS have a deficit in planning syllables.

In this study, no strong effects of syllable frequency on coarticulation and durational structure occurred in the utterances of the two groups. It is concluded that either the theoretical construct of a syllabary is false or the effect of syllable frequency manipulation were not strong enough in this study. As a last result it is found that the speech of children with DAS is much more variable than that of NS children. The higher within-subject variances are indications of a less automated process. Although studies on other speech disorders also report high variability, this finding supports the idea that children with DAS have a problem in automating speech more than normally speaking children.

In conclusion, the present study provides indications for a problem in the planning of syllables in speech production of children with DAS, in particular concerning prosodic aspects.

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CHAPTER FOUR

Evidence of motor programming deficits

Nijland, L., Maassen, B., & Van der Meulen, Sj. (in press). Evidence of motor programming deficits in children diagnosed with DAS. *Journal of Speech, Language, and Hearing Research*, to appear in April 2003.

ABSTRACT

In the present study the hypothesis of motor programming involvement in developmental apraxia of speech (DAS) is investigated by studying articulatory compensation. Five children with DAS and five normally speaking children (age 5;0 to 6;10 years), and six adult women produced utterances in a normal speaking condition and in a bite-block condition in which that the mandible was kept in a fixed position. Throughout the utterances, the course of the second formant was used to determine articulatory compensation and the effect of the bite-block on anticipatory coarticulation. Results showed, first, that the bite-block condition in normally speaking children, like in adult women, did not affect the extent of anticipatory coarticulation. In the speech of children with DAS large effects of the bite-block were found on vowel quality, which, contrary to expectations, had improved, and on coarticulatory patterns. These results are interpreted as a clear demonstration of deficient motor programming in DAS.

INTRODUCTION

The speech of children with Developmental Apraxia of Speech (DAS) is characterized by low intelligibility due to a large number of consonant errors, especially (contextual) substitutions and omissions. Researchers have been struggling to identify the exact speech symptoms of DAS as well as the accompanying non-speech characteristics, ever since Yoss and Darley (1974) first described the disorder as an impairment of motor programming and sequencing in children (see e.g., Davis, Jakielski, & Marquardt, 1998; McCabe, Rosenthal, & McLeod, 1998; Nijland et al., 2002; Velleman & Strand, 1994). Besides the many arguments raised in the debate on how to operationally define DAS, neuropsychological or neurolinguistic approaches have also advanced a diversity of views with regard to the underlying deficit. Explanations for DAS range from a disturbance localized at the level of phonological representation, the phonological encoding process, the generation of a phonetic program, up to the motor planning, programming and execution levels, and the generation and utilization of frames (Dodd & McCormack, 1995; Hall, Jordan, & Robin, 1993; Ozanne, 1995; Shriberg & Kwiatkowski, 1982; Stackhouse & Snowling, 1992; Van der Merwe, 1997; Velleman & Strand, 1994). There is a similar ongoing debate about acquired apraxia of speech in adults (for a review, see Ballard, Granier, & Robin, 2000; McNeil, Robin, & Schmidt, 1997) and developmental (limb) apraxia (Dewey, 1995). Although, as yet, no clear answer explaining the origin of DAS has been found, several models suggest that the location of the causal factor can be found somewhere in the transition from a phonological code into articulo-motor output, that is, phonetic planning, motor programming, or motor execution (e.g., Dodd & McCormack, 1995; Maassen, Nijland, & Van der Meulen, 2001; McNeil et al., 1997; Ozanne, 1995; Van der Merwe, 1997; Velleman & Strand, 1994).

The aim of the current study was not to find a clinical diagnostic marker to identify DAS. Rather, our study is a more hypothesis-driven evaluation to try and establish whether motor-programming deficits might be involved in DAS. In previous studies we tested whether the underlying deficit in DAS consists of deviant phonetic planning of speech. Indeed, evidence of a deviant use of syllable structure, suggesting a deviant phonetic *planning*, was found in a study in which we manipulated the syllable structure in an otherwise unchanging context (Maassen et al., 2001, Nijland, Maassen, & Van der Meulen, 1999, Nijland et al., 2003; Nijland & Maassen, in

press). In contrast to normally speaking children, children with DAS did not produce systematic duration changes as a function of imposed variations in the syllable structure, which was interpreted as a symptom of a phonetic planning deficit. Whether this deficit is segmental or prosodic in origin remains to be resolved.

Besides the main result that phonetic planning was disturbed in children with DAS, the high variability due to the inconsistency in repeated utterances and the slow speaking rate we found in the speech production of these children also suggested a motor *programming* deficit (Maassen et al., 2001, Nijland et al., 2003; Ozanne, 1995; Strand & McNeil, 1996; Weismer & Elbert, 1982). (during phonetic planning a phonological plan is translated into a phonetic plan, consisting of the spatial and temporal goals of the articulatory movements, whereas the precise articulatory instructions are specified during motor programming; Van der Merwe, 1997.) Also other speech characteristics, such as slow diadochokinetic rate and sound distortions, might indicate a motor programming deficit (Hall et al., 1993; Ozanne, 1995; Towne, 1994). Because the various studies did not provide an unambiguous answer as to whether disturbed phonetic planning or deficient motor programming of speech was involved in DAS, further research was deemed necessary. Therefore, in the present study, we investigated the possible involvement of a motor programming disorder in DAS. Lindblom, Lubker, and Gay (1979), among others, suggested that normal speech motor programming is ‘compensatory’, that is, motor programming allows ‘motor equivalence’ (Perkell, Matthies, Svirsky, & Jordan, 1993). One way of investigating motor equivalence is by studying speech production while a bite-block is clenched between the teeth, which method was adopted in the present study. It was our premise that the utterances produced in such a bite-block condition might reveal an underlying disruption in motor programming. Before elaborating on the hypotheses of the bite-block manipulation in the present study, we will first define motor programming and discuss various other bite-block studies.

Motor programming

Unlike the speech production model suggested by Levelt and Wheeldon (1994), in which they propose an articulatory network as the last stage of speech production in which the exact execution of the articulators is calculated, other speech production models (e.g., Ozanne, 1995; Van der Merwe, 1997) break up this final level into two stages: motor programming and motor execution. Motor programming is the stage in which the phonetic plans that characterize the spatial and temporal goals of the articulatory movements, the so-called ‘articulatory gestures’ (Browman & Goldstein, 1997) are translated into context-dependent motor specifications for the articulators, the so-called ‘coordinative structures’ (Browman & Goldstein, 1997), to be executed in the motor execution stage (Ozanne, 1995; Van der Merwe, 1997). This distinction between the specification of speech goals and the actual articulatory movements is supported by the observation that speakers can realize a given linguistic unit in many different ways dependent on physical abilities and contextual influences. For example, ‘lip closure’, which is one of the articulatory requirements when producing the consonant /p/, can be accomplished by movements of the mandibula, the lower lip, both lips, or combinations of these articulators. Varying speaking conditions, being, among other sources, the cause of variability in normal speech, require continuous adjustment of the speech movements. Most people can almost

without practice compensate for situations such as clenching a pipe between the teeth or wearing braces. Apparently, the motor system can immediately compensate for such changing circumstances (Guenther, 1995; Lindblom, Lubker, & Gay, 1979). Turning around this line of reasoning, we presume that in speech pathology a malfunction at the level of motor programming is likely to affect the extent of motor equivalence. Thus, the response of the articulatory system to perturbations is expected to provide interesting and useful information about the functional characteristics of speech motor programming.

Bite-block

Bite-block studies are used to investigate the process of compensation at the level of motor programming. Gracco and Abbs (1989) suggested two types of perturbations: early perturbation, which would yield so-called autogenic compensations indicative of an effective motor programming process, and later perturbation that would tap the stage of motor execution and reflect nonautogenic sensorimotor processes. Gracco and Abbs (1989) investigated the compensation to later perturbation by pulling down the participant's lower lip during speech production. Since we were interested in investigating deficits presumed to occur at the motor programming stage of speech production, that is, early perturbation, we opted for speech tasks executed with and without a bite-block.

Several studies investigated the compensation for a bite-block in normal and disturbed speech in adults and children, which resulted in divergent conclusions. Normally speaking adults generally demonstrate (almost) complete adaptation to articulatory perturbation, either immediately (Gay, Lindblom, & Lubker, 1981; Lindblom et al., 1979; Sussman, Fruchter, & Cable, 1995) or after an accommodation period (Baum, Kim, & Katz, 1997; McFarland & Baum, 1995). The results of these studies point to motor equivalence during speech production in adults, which means that adults are able to use different articulatory maneuvers to produce the same acoustic output. Lindblom et al. (1979) concluded that their findings of 'instantaneous' learning could not exclusively be attributed to similar past experiences, nor to invoking special motor mechanisms, but that they are the result of the compensatory character of normal speech motor programming, that is, being context-sensitive and predictive (Lindblom et al., 1979).

Whether children exhibit similar motor equivalence as observed in adults has been investigated in various studies. Whereas some researchers (e.g., Baum & Katz, 1988; De Jarnette, 1988; Smith, 1994; Smith & McLean-Muse, 1987a) concluded that normally speaking adults and children equally compensated for a bite-block, the results of other studies (e.g., Edwards, 1992) showed that children were not able to completely compensate for a bite-block. Edwards (1992) concluded that motor equivalence develops gradually in children. She interpreted the aberrant compensation patterns in children with phonological disorders as indicative of a problem in the speech motor processes. Furthermore, Edwards (1992) found deficient consonant production together with normal compensation in vowel production, which suggests that the speech motor processes involved in the production of consonants and vowels may be different. Investigating compensation for a bite-block in normally speaking children and children with a moderate articulation disorder, De Jarnette (1988) found no differences in motor equivalence across groups in the production of isolated vowels, possibly signifying that the vowel task was too simple,

which seems probable since also Towne (1994) and Rastatter, McGuire, and Blair (1987) reported a similar finding of normal motor equivalence in phonologically disordered children in relatively simple articulatory acts. These findings further emphasized the importance of studying consonant production as well as vowel production. Also studies in acquired apraxia of speech reported conflicting results; whereas several studies did not find effects of bite-block (e.g., Baum et al., 1997; Robin, Bean, & Folkins, 1989) others did (e.g., Sussman et al., 1995).

The inconsistent results of the bite-block studies might be due to differences in the parameters used in the data analyses that ranged from perceptual (e.g., Baum et al., 1997) and acoustic (e.g., Edwards, 1992) to kinematic variables (e.g., Robin et al., 1989; Smith & McLean-Muse, 1987b). Furthermore, the speech material investigated was quite diverse (consonants, plosives or fricatives, and vowels). In addition to these dissimilarities, the size of the bite-blocks varied across the studies, and they were not always custom-made. All these factors complicate a comparison of the various studies.

In the present study the shortcomings of the former studies, resulting in the above-mentioned miscellaneous results, were overcome as follows. First, we not only studied the compensation for bite-block in vowels, but also in consonants, both plosives and fricatives. Furthermore, the effect of bite-block on the anticipatory coarticulation of the target vowel on the preceding consonant and the preceding schwa was investigated. In this context, Hertrich and Ackermann (1999) showed that a lack of anticipatory coarticulation reflects programming deficits, but not execution problems. Second, since it would be difficult to compare the speech performance of children with DAS in the present study to the findings of other studies due to differences in research design, we decided to also examine the compensatory abilities in normally speaking children and adult speakers. Thus, we first looked at compensation in normally speaking children and compared this to the performance of the adults in order to assess whether children are able to compensate speech to the same extent as adults. Subsequently, we assessed compensation in children with DAS and compared this to compensation by normally speaking children. The extent to which children with DAS were able to compensate for the bite-block was used to analyze their speech motor programming. Aberrant patterns of compensation in children with DAS as compared to normally speaking children would thus be interpreted as evidence of a disturbance at the level of motor programming. A deficit at the level of motor programming was expected to result in less compensation in children with DAS.

Acoustic analyses, that is, second formant (F2) measurements, were used to assess the extent of compensation in the speech productions. Speaking with a bite-block necessitates that the tongue and lips compensate for the fixed, relatively low mandible to allow a correct production of speech sounds in which lip closure (e.g., consonant /b/) and elevation of the tongue tip (e.g., vowel /i/) or tongue body are required.

It was our first prediction that we would find an effect of bite-block on the stability of repeated productions. This would be manifested by an increase in variability of the F2-values in the bite-block condition. Secondly, since the speech task was relatively new to our participants, a less automated, more segmented type of speech production was anticipated, in which neighboring gestures would overlap less. As a consequence of this more segmented speech the utterances were expected to exhibit less anticipatory coarticulation, as measured in F2-values

(Nijland et al., 1999, Nijland et al., 2002). Finally, because it requires little or no compensation to produce speech sounds for which the jaw is already in the correct position, as was the case due to the use of the bite-block, an interaction effect was expected in which the difference in formant values between low and high vowels would be reduced in the bite-block condition as compared to the no-bite-block condition (Smith, 1987). Thus, the presumed underlying deficit in motor programming was expected to become manifest in an overall reduced ability to produce acoustically distinctive vowels, less anticipatory coarticulation and more variability.

Since, in this experiment, we confined ourselves to the characteristics of the speech production in children with DAS (and normally speaking children and adults), excluding children with other speech disorders, we will not be able to draw any conclusions about the specificity of the results for DAS. However, the aim of the present study was to demonstrate the involvement of motor programming deficits in a group of children who met the explicit diagnostic criteria for DAS. Reduced articulatory compensation in children with DAS compared to normally speaking children in a bite-block condition will be taken as evidence of a motor programming deficit.

METHOD

Participants

Participants were five children with DAS and five normally speaking (NS) children, between the ages of 5;0 and 6;10 years. All children were native speakers of Dutch. They had been selected from a group of 19 children with DAS (14 boys and 5 girls; age range: 4;11 – 6;10 years) and 19 NS children (matched for sex, age, and dialect) who participated in a more extensive project of which this study formed a part. The children with DAS, pupils of special schools for children with speech and language disorders, all met the clinical criteria described by Hall, Jordan, and Robin (1993) and Thoonen, Maassen, Wit, Gabreëls, and Schreuder (1996). The selection was based on samples of spontaneous speech, repetitive imitations of words and brief phrases, and a diadochokinetic task (using a Dutch Dyspraxia diagnosis test, the 'Dyspraxia Program'). The criteria the DAS group needed to meet comprised the following aspects: a complete phoneme repertoire with many phonemic errors, high frequency of consonant substitutions (and omissions in clusters), sequencing difficulties, inconsistent error patterns, and difficulty in producing complex phonemic sequences (Hall et al., 1993). Additional inclusion criteria were: no hearing problems, no language comprehension problems, no organic disorders in the orofacial area, no gross motor disturbances or dysarthria, and at least average nonverbal intelligence (Thoonen, Maassen, Gabreëls, & Schreuder, 1999; Thoonen, Maassen, Wit, Gabreëls, & Schreuder, 1996;). Table 1 lists, among other aspects, the characteristics on several (speech) tasks of the samples of children with DAS and NS children included in the present study. The maximum repetition rate (MRR) and substitution rates of the children thus selected as DAS met the criteria for diagnosing DAS according to the studies by Thoonen et al. (1999) and Maassen, Thoonen, and Boers (1997).

Table 1: Characteristics of the children with DAS and the NS children

Reynell																					
				Language		Schlichting language Production															
				comprehension		test					MRR		Substitution Meaningful utterances				Substitution nonsense utterances				
Group	Child	Age	sex	CQ	diff. age	SQ	diff. age	WQ	diff. age	mem.Q	Audio	Mono	tri-syll.	subcon	place	manner	voicing	subcon	Place	Manner	voicing
DAS	12	5-05	M	91	-0;08	76	-1;10	87	-0;09	104	<25 dB	3.02	0	12.18%	73.68%	42.11%	52.63%	43.94%	75.86%	41.38%	34.48%
DAS	13	5-06	F	84	-1;04	73	-2;02	72	-1;06	82	good	3.64	2.21	17.11%	69.23%	61.54%	23.08%	38.46%	68.00%	36.00%	56.00%
DAS	21	5-11	M	89	-0;10	73	-2;02	94	-0;09	96	good	3.20	0	7.75%	36.36%	45.45%	54.55%	-	-	-	-
DAS	26	6-06	F	84	-1;02	66	-3;05	79	-1;09	96	good	3.76	0	31.79%	75.00%	47.92%	27.08%	46.97%	64.52%	38.71%	64.52%
DAS	28	6-10	F	82	-1;08	66	-3;08	89	-1;07	89	good	4.23	3.61	21.58%	60.00%	43.33%	46.67%	39.39%	96.15%	38.46%	38.46%
Group		5-00		82		66		72		75		2.48	0	7.00%	18.00%	27.00%	8.00%	15.00%	30.00%	25.00%	30%
ranges		- 6-10		- 105		- 87		- 100		- 104		- 4.63	- 3.66	- 32.00%	- 80.00%	- 71.00%	- 100.00%	- 79.00%	- 96.00%	- 80.00%	- 83%
NS	36	5-00	M	-	-	-	-	-	-	-	good	4.85	3.68	0.72%	100.00%	100.00%	0.00%	16.67%	63.64%	63.64%	63.64%
NS	42	5-03	M	-	-	-	-	-	-	-	good	4.74	4.05	3.00%	0.00%	-	-	6.00%	25.00%	-	-
NS	49	5-11	M	-	-	-	-	-	-	-	good	4.97	3.06	0.65%	0.00%	100.00%	0.00%	12.50%	0.00%	16.67%	83.33%
NS	53	5-08	M	-	-	-	-	-	-	-	good	4.49	5.03	6.62%	33.33%	33.33%	33.33%	7.58%	20.00%	20.00%	80.00%
NS	58	5-06	M	-	-	-	-	-	-	-	good	4.44	5.08	2.07%	33.33%	33.33%	33.33%	9.09%	33.33%	0.00%	66.67%

Note. diff. age=age difference of the scores relative to norm-age scores; CQ=comprehension quotient (>80 is normal); SQ=sentence production quotient; WQ=word production quotient; Mem.Q=Auditory Memory Quotient; MRR=Maximum Repetition Task; monosyll=monosyllabic repetition (/pa/, /ta/, and /ka/), trisyll=trisyllabic repetition (/pataka/); Subcon=percentage consonant substitution; place/manner/voicing = percentage substitution of place/manner/voicing of subcon. Missing data are indicated with '-'. Group ranges for the DAS group are of the entire group of 19 children.

Not all children of the initial DAS group ($n=19$) were able to perform the bite-block task. Six children with DAS (compared to three children in the NS group) lacked the concentration and energy to participate in the bite-block condition, which was conducted after they had completed the other speech tasks, and another four children with DAS (compared to none of the NS children) were unable to clench the bite-block between the teeth while speaking¹. From the remaining group of nine children with DAS, five were selected for the analyses to be presented below. These five children also performed the speech tasks with bite-block at a second assessment conducted 16 months later. The upper and lower ranges of the scores of the total DAS group are also presented in Table 1 in order to show that these five children are representative of the total group (note that the five NS children, who were randomly selected, were all male, which is rather conservative for a control group, that is girls are assumed to show better performances on speech tasks than boys). The results of this sample of five children with DAS (and five NS children) will be presented below.

The results of six adult females were used as reference data. The women were between 20 and 30 years of age, and had no history of speech pathology, orthodontics, or hearing problems. We opted for women because, as compared to males, female voices and vocal tracts are more similar to those of children (Lee, Potamianos, & Narayanan, 1999).

Speech material

The speech material consisted of two-word utterances with simple CV-syllables, in which the relevant part was [əCV] and $C=/b,d,x,s/$ and $V=/a,i,u/$. These bisyllabic nonsense utterances were spoken within the carrier phrase 'hé d... weer' (/he d... wI:r/) "hey th... again" in a condition with a bite-block clenched between the teeth (bite-block condition) and in a normal speech condition (no bite block). This resulted in utterances like 'hé de ba weer' ("hey the ba again"), 'hé de di weer' ("hey the di again") and so on. The participants were asked to repeat the utterances after the experimenter. Each utterance type was elicited six times; the sequence of utterance types was randomized. This resulted in 72 utterances (i.e., 4 consonants * 3 vowels * 6 repetitions) per participant in each speaking condition (bite-block and normal speech condition). In case a child was not able to correctly produce an utterance (as judged by the experimenter immediately following the utterance), a second attempt was made straight away. This second (correct) production was subsequently used for the acoustic analysis.

A unidirectional dynamic microphone, mounted on a headset (Shure SM10A), and a tape-recorder (Kenwood KX54) were used to record the speech samples. The headset kept the microphone at a constant distance of 5 centimeters in front of the right corner of the subject's mouth.

The size of the bite-block, made of two-component AV-putty standard (Silagum ®), was about 1 x 1 x 0.5 cm (h x w x d). For each participant a separate bite-block was made (Netsell, 1985). To prevent the bite-block from slipping during task performance molar impressions were

¹ The 10 children with DAS who did not perform the bite-block task could not be defined as a clear subgroup of the total group by, for example, age (the youngest group) or percentage substitution (those with highest percentages of consonant substitution).

made by placing the bite-block between the participant's molars while it was solidifying. A spatula, 1 cm in height, clenched between the incisors, fixed the height of the bite-block. A string was attached to the bite-block to prevent it from being swallowed.

Acoustic analyses

The speech samples that were phonemically correct (as judged by the experimenter immediately during recording, see above, and informally checked by another experimenter during off-line analysis) were digitized at 25 kHz and the relevant sections, [əCV], were spliced out, using the Kay Elemetrics Computerized Speech Lab (CSL) analysis system. In these digitized utterances, the second formant (F2) trajectory was measured throughout the utterance (of schwa, consonant and second vowel). Information of the oscillogram, the FFT-spectrogram, and the energy window was used to determine the onset and offset of each segment, and markers were set at the segments' onsets and offsets at the plosive burst, using indications given by Nittrouer (1993, also see Nijland et al., 2002).

The formant values (with corresponding bandwidths) were obtained using pitch-synchronous LPC (triangular analysis window; 20 components autocorrelation with pre-emphasis of 0.950) analysis in the voiced sections of the signal (i.e., schwa and vowel), followed by the root-solving procedure (Buder, Kent, Kent, Milenkovic, & Workinger, 1996; Nittrouer, Studdert-Kennedy, & McGowan, 1989). For this, CSL automatically produced voice impulse markers (in the voiced sections of the signal) at the zero-crossings in the steep rises of the curve, corresponding with the closing of the glottis during voicing. Whenever necessary, the wrongly placed voice impulse markers (the automatic procedure in CSL was not always accurate) were corrected manually. In order to determine the formant values in the consonant, a separate LPC analysis was performed on a Hamming window of 20 ms. The measurement window in the fricatives (/s/ and /x/) was centered at 20 ms before offset; in the plosives (/b/ and /d/) it was centered at the middle of the plosive burst. Thus, the F2 values were obtained at 6 locations in the utterance: at schwa-midpoint ('mid-schwa') and schwa-offset ('end schwa'), in the consonant (20 ms before offset in the /s/ and /x/; at plosive burst in the /b/ and /d/; 'consonant'), at the vowel transition onset ('transition onset') and transition end ('transition end'), and at vowel midpoint ('mid V').

Although F2 values can be used in the comparison between the bite-block condition and the normal speech condition within one speaker, physiological differences between speakers prevent these F2 values from being used directly to evaluate differences between individual participants. In order to correct for systematic differences among speakers, that is, as a means of speaker normalization, F2 ratios were calculated for each speaker individually. This was done by dividing the mean F2 values (of 6 repetitions) in /i/-utterances by the mean F2 values in /u/-utterances. Subsequently, these i/u-ratios can be interpreted as an index of the distinction between the utterances: a ratio of 1 means that the formant frequencies of both /i/ and /u/ utterances are equal, whereas the higher the ratio is above 1 the more distinction there is between the utterances.

Statistical analyses

The F2 values were used to statistically test the effect of bite-block within each participant, for which the statistical package SPSS for Windows version 9.0.1 was used. The main effect of bite-block was tested by using Paired-Samples *t*-tests comparing the mean F2 values (average of 6 repetitions) of the two speaking conditions (bite-block and normal speech condition) across utterance types. Bonferroni's correction was adopted to correct for multiple comparisons (Winer, Brown, & Michels, 1991).

In order to test interaction effects of bite-block with the utterance type, that is, Consonant and Vowel ², analyses of variance were conducted with Bite-block, Consonant, and Vowel as within-subject factors. This was done for the three groups separately in order not to violate the assumption of homogeneity (Winer et al., 1991).

Subsequently, the variability in measured F2 values was determined among speakers (between-subject variance), between utterance types, and in repetitions of utterances (within-subject variance) in the three groups and both speaking conditions separately. The effect of bite-block on the variability within each group and differences in variability among groups was tested by calculating the F-values (i.e., by dividing Mean Squares). The significance level was corrected for multiple comparisons using Bonferroni's correction.

To evaluate group differences F2 ratios were used. The effects of Bite-block, Consonant and Vowel among the groups were tested on the F2 ratios using analyses of variance. For this, the NS children were compared to the adult women (AW), and the children with DAS were compared to the NS children. The groups were split in NS versus AW and DAS versus NS in order to be able to test the significance of an interaction effect of bite-block with group.

RESULTS AND DISCUSSION

In this section we will first discuss the effect of bite-block on the F2 values in the three groups separately (adult women, NS children, and children with DAS). Subsequently, the variability in formant values measured within and across the three groups is given, allowing us to evaluate whether production with a bite-block induced more variability and whether the groups differed in this aspect. Finally, the effects of bite-block and coarticulation across the groups will be discussed on the basis of the F2 ratios.

Bite-block effect within groups

The effect of the bite-block on the second formant (F2) throughout the utterances was tested in each group. Kolmogorov-Smirnov test for normality showed that the F2 values were normally distributed (all *Z*'s (72) < 1.32; ns.), thus permitting the use of a *t*-test. The mean F2 values of the adult women at all measurement points were not significantly affected by Bite-block (all *t*'s (71) < 1.79; ns.). In the NS children the F2 values were generally higher due to the bite-block; this

² The notation Vowel will be used to refer to the factor in the analysis of variance as opposed to the measurement points ('mid schwa' and 'end schwa' in the schwa, and 'transition onset', 'transition end' and 'mid V' in the vowel).

increase was significant in the schwa ('mid schwa': $t(58)=2.85$, $p<0.01$; 'end schwa': $t(58)=3.49$, $p<0.001$). In the children with DAS the bite-block led to lower mean F2 values, which was significant in both schwa and vowel ('mid schwa': $t(48)=2.50$; 'transition onset': $t(52)=2.38$; 'mid V': $t(55)=3.10$, $p<0.01$).

Thus, the adult women were able to compensate for the bite-block, whereas both groups of children proved unable to do so completely. However, the bite-block manipulation had a differential effect on the two groups of children. Reverting to the hypothesis that problems in motor programming will appear in aberrant patterns of compensation in the children with DAS as compared to the NS children, we interpret the difference in compensation between the NS children (higher F2 values) and the children with DAS (lower F2 values) as a first indication of a disturbance at the level of motor programming. Furthermore, results of an earlier study (Nijland et al., 2002) showed that in the normal speech condition children with DAS had higher F2 values than NS children in the same utterances. Thus, as the children with DAS showed lower F2 values as a consequence of the bite-block, this suggests that the children with DAS were actually helped by the bite-block to produce F2 values that were more similar to those of the NS children. This, furthermore, indicates that children with DAS have a disturbance at the level of motor programming in the normal speech condition, which is to a certain extent corrected for in the bite-block condition. Other differential findings for the two groups will be discussed later in this section.

In order to investigate whether Consonant and Vowel influenced the extent of the bite-block effect, that is, whether particular CV combinations led to larger effects of the bite-block than others, analyses of variance were conducted for each group separately. Although no main effect of Bite-Block was found in F2 values of the adult women, one significant interaction effect of Vowel with Bite-block was found at 'transition onset' ($F(2,10)=5.04$, $p<0.05$), in which the effect of Bite-block was stronger in the vowels /i/ and /u/ than in /a/.

In the NS children significant interaction effects of Vowel with Bite-block were found at the measurement points 'consonant' ($F(2,8)=9.86$, $p<0.01$) and 'transition onset' ($F(2,8)=4.62$, $p<0.05$), where the effect of Bite-block was stronger in the utterances with /i/ and /u/ than in the utterances with /a/. Furthermore, at 'consonant' it was found that /bi/ and /bu/ showed the largest effects of the bite-block, as indicated by a significant interaction effect of Consonant with Vowel with Bite-block ($F(6,18)=6.99$, $p<0.01$).

The children with DAS showed a significant interaction effect of Bite-block with Vowel at 'end schwa' ($F(2,7)=6.25$, $p<0.05$), that is, the decrease in F2 value due to the bite-block was strongest in /i/-utterances. Interaction effects of Bite-block with Consonant and Vowel (CV combination) were significant at 'mid schwa' ($F(6,15)=3.20$, $p<0.05$), 'consonant' ($F(6,17)=4.07$; $p<0.01$), and 'transition onset' ($F(6,18)=3.17$, $p<0.05$). At 'mid schwa' and 'consonant' the interaction effect was manifested by a stronger decrease in F2 values due to the bite-block in the utterances with /ba/, /bi/, /bu/, /xa/, and /xi/ compared to those for the other utterances. At 'transition onset' a decrease in F2 values was visible in the utterances with /ba/, /bi/, /bu/, /xa/ and /su/.

To summarize these effects, it was shown that interaction effects of bite-block with consonant-vowel combination occurred hardly in the adult women and mostly in the children

with DAS. First, in all three groups the interaction effects of Bite-block with Vowel showed that the utterances with /i/ and /u/ were more affected by the bite-block than the utterances with /a/. This significant interaction effect extended from ‘transition onset’ in the adult women to ‘end schwa’ in the children with DAS. Evidently, compensation for the bite-block is easier for the open vowel /a/ than for the more closed vowels /i/ and /u/. Second, the significant interaction effects of Bite-block with Consonant and Vowel showed that in the NS children the utterances with /bi/ and /bu/ were more affected by the bite-block than the other utterances. In the children with DAS a relatively large effect was found for the utterances with /ba/, /xa/, /xi/ and /su/. Thus, not being able to move the mandible disturbed the speech of the children with DAS more and at an earlier stage in the utterance than it did the speech of the NS children. Furthermore, it was apparently harder to completely compensate for the bite-block in the utterances with /b/ and /x/ than in the other utterances.

Variability

In earlier studies it was found that the within-subject variability was higher in children with DAS than in NS children (Nijland et al., 1999, Nijland et al., 2002; 2003) as was also found in acquired apraxia (Strand & McNeil, 1996). In the present study, the variances of formant values that were due to repetition of the same utterances (within-speaker variability), speaker variability (between-subject variability), and utterance type were calculated³. The effect of bite-block on these variances was tested in each group separately, and compared across groups. These analyses addressed the following questions: Does the bite-block affect the stability of the utterance production? And, if so, is this effect larger in the children as compared to the adult women, and is it larger in the children with DAS as compared to the NS children?

Figure 1 displays the within-subject standard deviations of F2 values for each group and speaking condition. Significant effects of bite-block are indicated with an asterisk. Figure 1 shows a significant decrease of variability in F2 values due to bite-block in the adult women (AW) at ‘mid schwa’ and ‘end schwa’ ($F(360,369)=2.04$ and $F(340,345)=1.54$, $p<0.05$, respectively), and a significant increase of variability at ‘consonant’ ($F(266,278)=1.56$, $p<0.05$). The within-subject variance in the NS children did not change significantly as a result of the bite-block (all $F_s(316,332)<1.26$, ns.). In the children with DAS, the within-subject variance in F2 values significantly increased due to the bite-block at the first four measurement points (all $F_s(229,215)>1.55$, $p<0.05$). In contrast, the bite-block did not affect the variability in F2 values among speakers and between utterance types.

Thus, the adult women showed a reduction in within-subject variability measured in the schwa, which indicates an increase in stability due to the bite-block. However, in the consonant the variability across repetitions increased. This finding suggests, in accordance with Edwards (1992), that the speech motor processes of consonants and vowels may be different. Whereas in

³ Some studies recommend to use coefficients of variation, that is the ratio of standard deviation over the mean, rather than standard deviations because of a possible correlation between sd and mean (see e.g., Lee et al., 1999). However, since in the present data there was no strong correlation between the two (correlations were smaller than 0.195), standard deviations (i.e., the square root of variance) were used.

the NS children no significant effect of bite-block was found in the variability of repetitions, in the children with DAS the bite-block reduced the stability of the productions across repetitions.

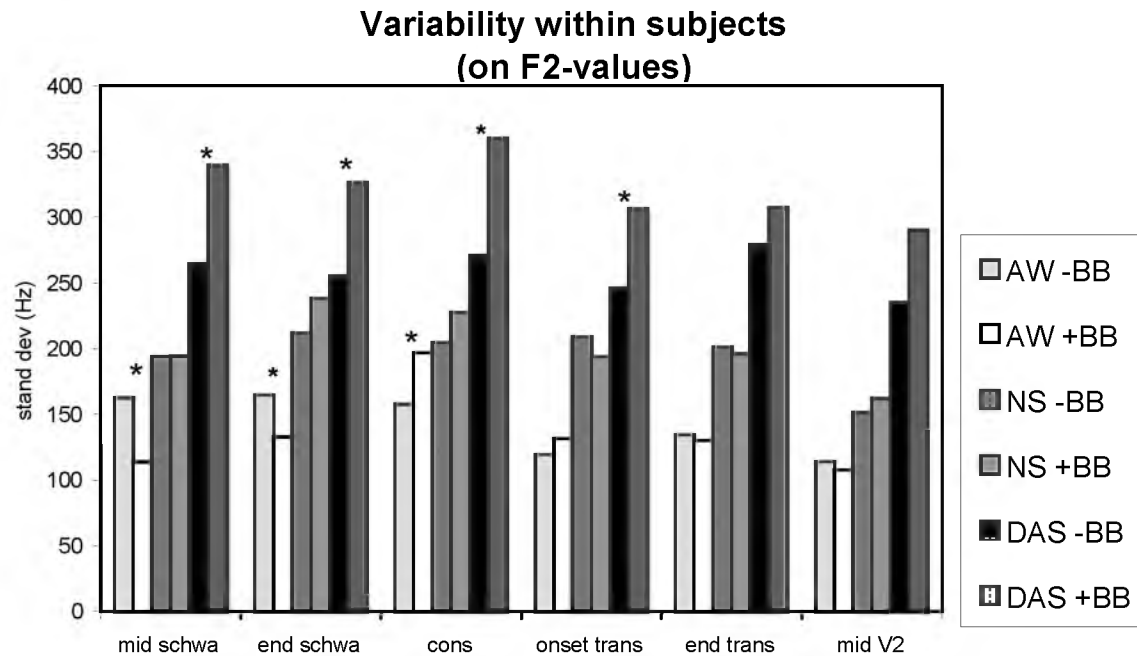


Figure 1. Within-subject variability: standard deviations of F2 values measured at 6 locations in the utterances of adult women (AW), normally speaking children (NS) and children with DAS, spoken in both the normal speech condition (-BB) and the bite-block condition (+BB).

A comparison of the within-subject variability across the three groups resulted in significant differences. The NS children showed larger within-subject variability than the adult women in both speaking conditions at all locations (all $F(316,357) > 1.65$, $p < 0.05$), except 'mid schwa' in the normal speech condition and at 'consonant' in the bite-block condition. Furthermore, the children with DAS had larger variances in the normal speech condition than the NS children, variances being significantly larger at four out of six locations (all $F(171,270) > 1.75$, $p < 0.05$) though not significant at 'end schwa' and 'transition onset'. In the bite-block condition, at all locations the within-subject variability in F2 values was significantly larger in the children with DAS as compared to the NS children (all $F(154,232) > 1.88$, $p < 0.05$).

In conclusion, the results on the variability analyses revealed that within-subject variability was smallest in the adult women and largest in the children with DAS. In the NS children the bite-block did not change the variability, whereas the children with DAS did show a resultant increase of variability. Since the bite-block caused the F2 values to decrease in the children with DAS, an increase in within-subject variability could, thus, not be explained by higher F2 values. The same applies to the comparison across groups, where the children with DAS displayed significantly higher within-subject variability as compared to the NS children in both speaking conditions, but the difference in F2 values decreased due to the bite-block. Together, these results provide another indication that the children with DAS could not compensate for the bite-

block like the NS children did, which corroborates the hypothesis of a disturbance at the level of motor programming.

Formant ratios

Systematic differences between participants due to anatomical variability complicated the comparison of formant values across groups. To facilitate a more valid comparison, F2 ratios were calculated for each speaker individually to allow speaker normalization. To this end, the mean F2 values (average of 6 repetitions) throughout the /i/-utterances were divided by the mean F2 values throughout the /u/-utterances. Besides allowing a comparison of individual speakers, the F2 ratios provided a measure to distinguish the utterances: high F2 ratios in the second vowel indicating large distinctions between vowels, and, in the measurement points preceding the vowel, reflecting the coarticulation effect of the upcoming vowel.

Figure 2 displays the F2 ratio patterns of the adult women (2a), the NS children (2b), and the children with DAS (2c), respectively. The dotted lines represent the ratios in the normal speech condition; the solid lines those in the bite-block condition. High F2 ratios at the location 'mid V' indicate that there is a large distinction between vowels. Of course, high ratios here were expected, demonstrating that vowels are characterized by different F2s. Earlier in the utterance the ratios indicate the extent of anticipatory coarticulation of the second vowel on the preceding consonant and schwa: the higher the ratios, the greater the anticipatory coarticulation.

Analyses of variance were conducted on the F2 ratio data in order to compare the different groups (the F2 ratios were normally distributed across the groups: all Kolmogorov-Smirnov Z's < 1.23; ns.). In order to find significant differences of bite-block effect between the groups, the NS children were compared to the adult women and the children with DAS were compared to the NS children (post-hoc analyses of Bite-block*Group were not possible). Table 2 (upper half) shows the mean F2 ratios with standard deviations of the three groups in the two speaking conditions. Missing values arose in the /u/-utterances (also see Nijland et al., 2002) in the bite-block condition (especially in the group with DAS), causing the lower number of cases. The results of the analysis of variance are given in the lower half of Table 2.

First, the NS children were compared with the adult women (AW) (Table 2; also see Figures 2a and 2b). The groups did not differ significantly in the two speaking conditions, except for one location in the bite-block condition, that is, 'transition end'. The absence of significant effects of bite-block with group indicates that the coarticulation in the utterances of the adult women and the NS children was similar in both the bite-block and normal speech condition.

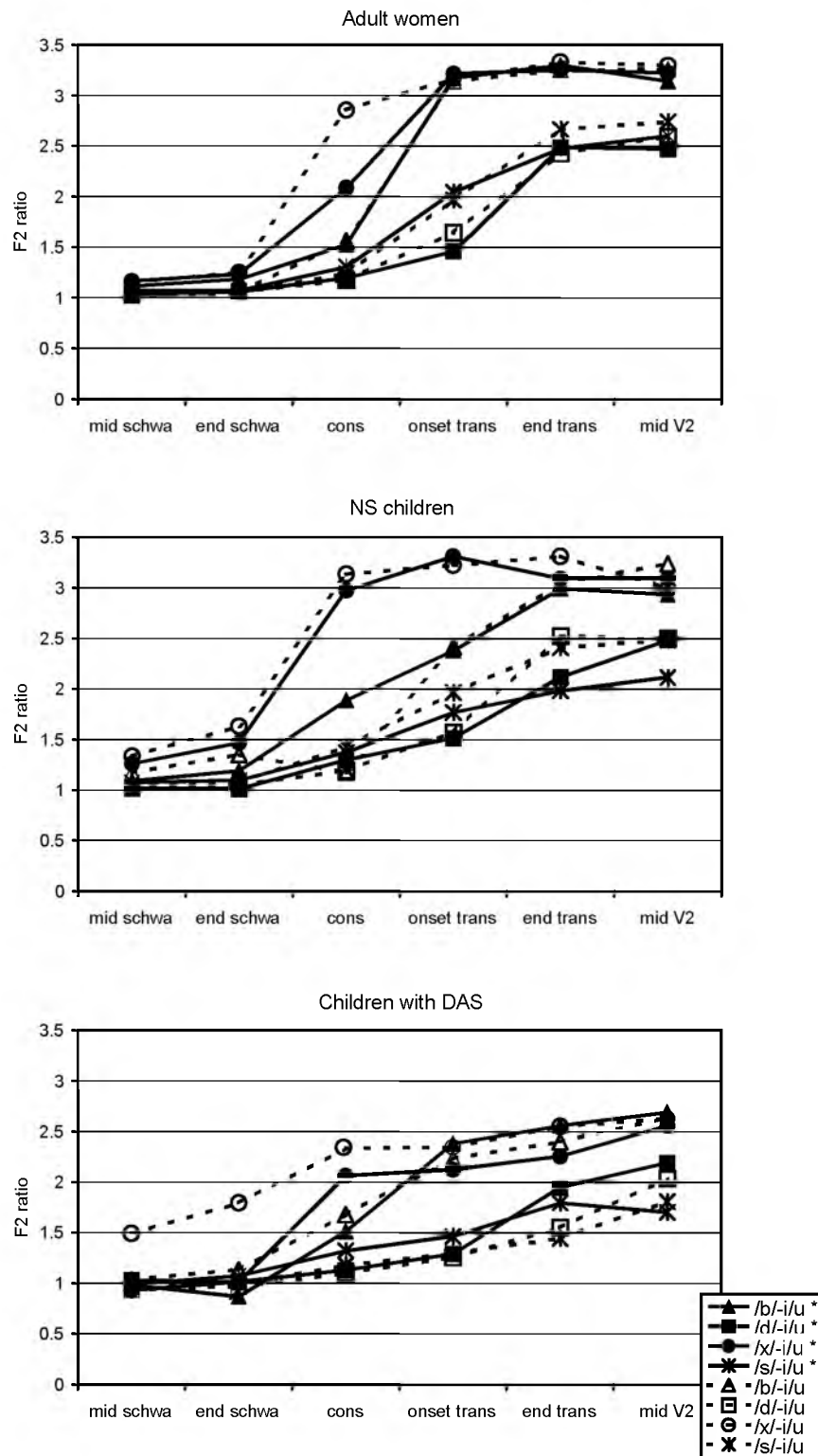


Figure 2. F2 ratios (i/u) of adult women (a), normally speaking (NS) children (b), and children with DAS (c) without bite-block (broken lines) and with bite-block (solid lines).

Table 2. Descriptive data and analyses of variance on the F2 ratios of the Adult Women (AW), normally speaking children (NS) and children with DAS. (N = Number of cases; M = Mean F2 ratio; sd = standard deviation.)

		Descriptive Statistics																	
Group	Cond.	Mid schwa			end schwa			Cons			Transition onset			transition end		Mid V			
		N	M	sd	N	M	sd	N	M	sd	N	M	sd	M	sd	N	M	sd	
AW	- BB	24	1.07	0.10	23	1.11	0.13	19	1.85	0.83	19	2.67	0.73	20	3.09	0.56	24	3.01	0.43
	+ BB	24	1.10	0.09	23	1.14	0.12	19	1.74	0.71	19	2.59	0.83	20	3.03	0.58	24	2.88	0.43
NS	- BB	19	1.15	0.14	19	1.26	0.26	16	1.76	0.86	19	2.41	0.80	17	2.82	0.59	20	2.83	0.47
	+ BB	19	1.13	0.12	19	1.22	0.20	16	2.00	0.79	19	2.24	0.80	17	2.60	0.59	20	2.67	0.45
DAS	- BB	16	1.14	0.28	15	1.18	0.42	15	1.54	0.68	16	1.76	0.65	16	2.02	0.65	19	2.25	0.58
	+ BB	16	1.04	0.15	15	1.00	0.14	15	1.48	0.56	16	1.93	0.71	16	2.16	0.62	19	2.49	0.75

ANOVA							
Group	Factor	mid schwa	end schwa	Cons	transition onset	transition end	mid V
AW	BB	1.53	1.61	.71	.68	.36	3.95
NS	BB	.33	.85	2.43	2.84	2.82	2.92
DAS	BB	1.61	2.54	.18	3.50	2.87	8.61 **
Comparison	G (–BB)	3.78	5.85	.03	.65	.86	1.75
AW-NS	G (+BB)	.88	2.40	1.65	3.06	4.60 *	2.35
	BB	.11	.00	.47	3.20	2.93	6.79 **
	BB*G	1.54	2.31	3.11	.42	.93	.04
Comparison	G (–BB)	.06	.80	.79	8.74 **	17.96 ***	12.04 **
NS-DAS	G (+BB)	4.30 *	12.80 **	4.77 *	1.44	4.20	.82
	BB	2.20	3.91	.60	.00	.21	.64
	BB*G	1.17	1.60	1.94	6.04 *	5.32 *	10.63 *

Note. * $p < 0.05$; ** $p < 0.01$; *** $p < 0.001$; Analyses of variance: Effect of bite-block (BB) for each group; Effect of group (G) in both speaking conditions (normal speech condition – BB; and bite-block condition +BB), main effect of bite-block (BB), and the interaction effect of bite-block and group (BB * G).

Second, the F2 ratios of the children with DAS (Figure 2c) were compared to those of NS children (Figure 2b). The significant differences between the groups are shown in the analyses of variance in Table 2. The F2 ratio curves and the effect of bite-block (Table 2: BB) in each group separately show that the children with DAS reacted differently to the bite-block than the NS children did, as was also found in the F2 value analyses. Overall the F2 ratios decreased in the NS children due to the bite-block, although not significantly, whereas in the children with DAS the ratios increased in the second vowel (significant at ‘mid V’).

Furthermore, in both speaking conditions the F2 ratios of the NS children overall appeared to be higher than the F2 ratios in the children with DAS. This means that the children with DAS apparently make less distinction between the vowels (at ‘mid V’) and have overall less anticipatory coarticulation as compared to the NS children. The results of the analyses of variance, in Table 2, show that, in the normal speech condition, the two groups differed significantly at the end of the utterance (‘transition onset’ to ‘mid V’). In the bite-block condition the difference in F2 ratios

was significant in the schwa and consonant. Thus, due to the bite-block, the difference between the NS children and the children with DAS disappeared at 'transition onset', 'transition end' and 'mid V', but they became larger in the schwa and consonant (this is statistically confirmed in the interaction effect of bite-block with group, Table 2: BB*G).

In conclusion, although the bite-block enabled the children with DAS to articulate the various vowels better, they still showed aberrant coarticulation patterns (i.e., less anticipatory coarticulation) as compared to the NS children. This finding contradicts the view that children with DAS show diminished anticipatory coarticulation merely because they make less distinction between the vowels. Evidently, the smaller coarticulation effect we found in the children with DAS, both in the present study and our former studies (Nijland et al., 2002), originates from another source.

GENERAL DISCUSSION

In this study we addressed the question whether children with DAS show a disturbance at the motor programming stage of speech production. As it was assumed that a bite-block would intervene at this stage of speech production, we evaluated the characteristics of motor programming in children with DAS by investigating their compensatory abilities to a bite-block manipulation and compared these with the performance results of normally speaking (NS) children. Prior to this, we weighed the compensatory abilities of the NS children against those of normally speaking adult women in order to collect reference data. It was argued that if the children with DAS would show the same effects in the bite-block condition as the NS children, their compensatory abilities could be considered similar, which would rule out a deficit at the motor programming level. Conversely, if the bite-block condition would reveal different effects for the two groups, this could then be seen as indicative of disturbed motor programming. Note that this assumption does not imply that we exclude the possibility of concurrent, additional problems at other levels of the speech production process. However, in the present study we restricted our investigations to the motor programming stage. The results showed that, although the NS children, as compared to the adult women, were not able to completely compensate for the bite-block (the F2 values increased), the children with DAS more clearly exhibited compensation problems and, moreover, reacted differently to the bite-block manipulation than the NS children. We therefore conclude that the findings on the speech production of the children with DAS investigated in the present study are indeed indicative of a problem at the level of motor programming.

Next, we would like to draw attention to several other noteworthy results of our study. First, the present finding that the adult women did not show an effect of the bite-block manipulation corroborates the results of earlier studies that had found evidence of immediate compensation (Baum & Katz, 1988; Lindblom et al., 1979) or compensation after an accommodation period (Baum et al., 1997; McFarland & Baum, 1995). Although the bite-block resulted in significantly higher formant values in the schwa in the NS children, the F2 ratios did not reveal significant differences between the NS children and the adult women for either of the two conditions. This means that the extent of their anticipatory coarticulation was not affected in the bite-block

condition. It needs to be noted, however, that due to the anatomical differences between adult women and children (women having a larger facial skeleton), the use of the same size bite-block might have posed a greater obstacle for both the NS and DAS group, which may account for the few differences observed between these two groups.

Second, as was the case with the NS group, the children with DAS also could not completely compensate for the bite-block, which was demonstrated by the lower F2 values in the schwa, at the onset of the transition in the vowel, and at 'vowel midpoint'. However, in contrast to the NS children, in the DAS group a bite-block effect did emerge in the F2 ratios, namely in an increase of F2 ratios at 'vowel midpoint'. As a consequence, at 'vowel midpoint' the difference between the two groups of children was neutralized. Nevertheless, the group difference with respect to coarticulatory patterns was still considerable in that the children with DAS showed less anticipatory coarticulation throughout the utterances. Thus, although the children with DAS were able to enhance their articulation of the vowels, which led to more 'normal' F2 ratios, the coarticulation patterns were still aberrant.

The finding that the children with DAS appeared to benefit from the bite-block was quite remarkable. We had expected that speaking with a bite-block would increase their speech problems, but found the opposite to be true. A similar pattern was described by Netsell (1985) in an example of a subject with Parkinson's disease. He speculated that the bite-block both slowed the speaking rate and required the subject to increase the range of lip and tongue movements. An alternative explanation might be that the reduction of degrees of freedom (jaw movement is fixed) may result in an improvement of the lip and tongue movements. On the other hand, the results of the analysis of variability in repeated utterances (within-subject variance) showed an increase in variability measured in F2 values in the children with DAS due to the bite-block, which was not found in the NS children. This indicates that the children with DAS, although improving the quality of their output, did experience difficulty in the articulatory process while speaking with a bite-block. Furthermore, the finding of less anticipatory coarticulation in combination with improved vowel articulation is paradoxical. Since coarticulatory effects are proportional to the distinctiveness of the sounds producing the effects, a greater distinction between two targets may be expected to produce greater coarticulatory effects earlier in the utterance. Hypothetically, weaker anticipatory coarticulation might be due to longer time intervals in DAS as compared to NS children. Recent analyses of durational patterns showed that segment durations of children with DAS are longer than those of NS children. However, in both present groups the bite-block condition resulted in longer segment durations (submitted paper by Nijland, Maassen, & Schreuder, 2002). Since the NS children did not show a decrease in anticipatory coarticulation due to the bite-block, slower speaking rate does not seem to be the only explanation. There seems to be another mechanism at play preventing the improved vowel production from having an effect on the coarticulation in children with DAS.

From the results of improved vowel quality, less stability in repeated utterances and less anticipatory coarticulation, we infer that the children with DAS experienced problems at the motor programming level. Thus, it is our conclusion that the much debated underlying deficit in children with DAS (among others Dodd & McCormack, 1995; Ozanne, 1995; Van der Merwe, 1997; Velleman & Strand, 1994) may indeed be a disturbed motor programming.

Thirdly, since the aim of the present study was not to find a diagnostic marker for DAS, but merely to investigate whether or not motor programming problems might be involved in DAS, we exclusively studied the speech disorder DAS. As a consequence, no conclusions can be drawn as to the specificity of this result for DAS as compared to other speech disorders. Several studies found normal motor equivalence in children with phonological disorders (De Jarnette, 1988; Rastatter, McGuire, & Blair, 1987; Towne, 1994). It is therefore probable that effects similar to those observed in the present study might be found in children with other articulation disorders (e.g., dysarthria). In the future, studies comparing DAS and other speech disorders are recommended.

Fourth, the interaction effect of bite-block with the target vowel that was found in the F2 values of all three groups studied here corroborates the findings of Baum et al. (1997). They suggested that the high vowels /i/ and /u/ would be affected more by a large bite-block than the vowel /a/. It needs to be noted that this effect was more pronounced and occurred earlier in the utterances of the children as compared to those of the adult women. Additionally, Baum et al. (1997) showed that fricatives were more vulnerable to distortion when a bite-block was used than the plosive [t]. In the present study, the interaction effect of bite-block with vowel with consonant failed to confirm the finding of Baum et al. (1997) since the utterances with /bi/, and /bu/ in the NS children were more affected than the other utterances. In the children with DAS also utterances with /x/ were more affected by the bite-block than those with /d/ and /s/. Apparently, rather than being associated with a difference in manner of articulation (fricatives versus plosives), the vulnerability of consonants to the bite-block seems more connected to a difference in place of articulation, which is similar for /d/ and /s/ but different for /b/ and /x/ (also see the discussion on homorganic versus heterorganic articulation by Hardcastle & Edwards, 1992; Nijland et al., 2002; Nitttrouer, 1993).

In conclusion, the results of the present study show that the group of children with DAS participating in our experiment displayed different coarticulatory patterns than the NS children. Moreover, they exhibited more variability within repeated utterances than the NS children and the adult women, both in the normal and in the bite-block condition. This indicates that the children with DAS were not able to compensate for the bite-block to the same extent as the NS children managed to do. Thus, although we by no means exclude the possibility of concurrent problems at other levels of the speech production process, in our opinion the present study clearly demonstrates a problem in motor programming in DAS.

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CHAPTER FIVE

Durational patterns reflecting deviant durational control

Nijland, L., Maassen, B., & Van der Meulen, Sj. (2002). Durational patterns reflecting deviant durational control in children with DAS. *Manuscript submitted for publication.*

ABSTRACT

The aim of the present study is to investigate durational control in children with developmental apraxia of speech in order to answer the question whether the slow speaking rate as found in these children reflects the underlying disorder or can be considered a compensatory strategy. Two experiments were conducted in which (1) the durational patterns, that is intrinsic and relative durational characteristics due to contextual interdependency and (2) compensatory strategies to an experimentally induced problem, that is a bite-block, are compared between children with DAS and normally speaking (NS) children. Results of Experiment 1 showed longer segment durations, a lack of significant contextual interdependency, and higher variability in children with DAS as compared to NS children, which suggested that durational control was less strict. Experiment 2 corroborated the suggestion of deviant durational control as the underlying problem in the longer durations as found in children with DAS.

INTRODUCTION

In this paper we present a study on the issue of durational control of speech in children with developmental apraxia of speech (DAS). DAS is a speech disorder that emerges during development and is primarily characterized by low intelligibility due to a large number of consonant errors, especially (contextual) substitutions and omissions. In the research of articulation disorders one struggles to distinguish compensatory strategies from the direct effects of the speech disorders. Is a symptom the result of normal behavior or can it only be explained as the result of deviant behavior? The first can be understood as a compensatory symptom whereas the latter can be explained as a symptom of the disorder (Hendriks & Kolk, 1997). Whether the slow speaking rate and long segment durations as found in DAS should be considered a result of the disorder or an effect of compensation will be discussed in this paper.

Clinical descriptions of DAS mention durational characteristics. The speech of children with DAS tends to be (a) *slower* than speech of normally speaking age-mates, but differences in (b) durational *pattern* also have been described. First, among the characteristics of DAS referring to overall long duration are: monotone and slow rate of speech (Guyette & Diedrich, 1981; Ozanne, 1995), increased sentence duration and consonant and vowel durations (Hall, Jordan, & Robin, 1993), which is also visible in slow diadochokinetic rate (e.g. McCabe, Rosenthal, & McLeod, 1998; Strand, 2001). These durational characteristics are not unique for DAS. Slow rate is one of the most generally observed features of speech motor disorders, which is observed in several types of dysarthria (like e.g. spastic and ataxic dysarthria, Hertrich & Ackermann, 1999), as well as 'higher level' motor speech disorders such as apraxia of speech and DAS. This implies that overall rate is a very relevant characteristic of speech disorders, but that its differential diagnostic value is extremely limited.

Second, differences between children with DAS and normally speaking (NS) children have been found in the intrinsic and relative durational characteristics of their speech, which in the present study will be termed as 'durational pattern'. The durational patterns are especially affected by segmental and suprasegmental context. Characteristics that are reported are: scanned speech (Kent, 2000), equal stress (unstressed syllables are stressed, Velleman & Shriberg, 1999), and prolongations of transitions and steady states (Yoss & Darley, 1974). Shriberg et al. (1997a)

interpreted the deviating durational patterns in the utterances of children with DAS as resulting from problems in rhythm and prosody, which correspond to durational control, rather than reflecting differences in movement durations (or 'prearticulatory sequencing'). As Manuel (1999) said: '... prosody clearly has to do with timing, so it seems likely that prosody and temporal coordination of articulatory gestures are strongly linked' (p.196). Also possibly related to durational pattern are voicing errors, in which the temporal coordination of physiological speech mechanisms is disrupted as for instance in discoordination of voice onset time (e.g., Itoh et al., 1982), and errors in which vowels are substituted for diphthongs or vice versa (Pollock & Hall, 1991). Furthermore, insufficient temporal coordination for appropriate voicing control affects a neighboring consonant that depends on the voicing status of the (incorrectly produced) phonetic context (Velleman & Strand, 1994).

Underlying the longer durations two different mechanisms might be operative. First, the longer durations may be a direct, primary effect of the speech disorder. Thus, if the movements of the articulators are impeded due to motor problems, the direct consequence is slower movements and longer durations. Alternatively, the longer durations may result from a compensatory strategy. According to this interpretation slowing down the process of speech production is used as a strategy to adapt to the speech disorder in order to avoid phonetic errors (Yoss & Darley, 1974).

Apraxia of speech (AOS), an acquired speech disorder in adults to which DAS is often compared because of the similarity in characteristics, also shows slow speaking, long segment durations (Kent & Rosenbek, 1983), and deviant durational patterns such as no normal linguistic effect of vowel duration lengthening in syllable final position and stressed syllables (Caligiuri & Till, 1983), nor shortening duration in sentence context relative to word context (Strand & McNeil, 1996). In contrast, Weismer and Fennell (1985) found that, although absolute durational measures were aberrant in patients with neurogenic speech disorders, their relative timing across speaking rates was stable. However, this result has to be taken with cautious since the group of patients included varying neurogenic disorders. Studies showing that apraxic speakers were not able to increase their speaking rate, suggest that slowness in apraxia is a primary, not a compensatory symptom (Rosenbek & McNeil, 1991; Skenes, 1987). Rosenbek and Wertz (1983) also suggest that deviant timing expresses the nature of the disorder and the durational changes are not compensatory. In contrast, an example of durational alternations as compensatory effects is suggested by Hertrich and Ackerman (1995). They found a disproportionate lengthening of inter-word pauses during slowed speech by normal speakers. These authors conclude that speakers are able to switch to another speaking mode, resulting in a different durational pattern.

The focus of the present study is to investigate the durational pattern and compensatory behavior in order to explain the processes of durational control (either normal or deviant). Stability of durational pattern between varying phonetic contexts and speech conditions indicates that durational control processes are involved. (Note that this study does not attempt to make a contribution to differential diagnosis.)

The present study is subdivided in two experiments. Experiment 1 dealt with the question whether, apart from the longer durations in the speech of children with DAS, differences in durational *pattern* also may be found between children with DAS and NS children. The second

experiment further investigated whether longer segment durations could be interpreted as a direct consequence of the disorder or as a compensatory strategy. For this, compensatory strategies to an experimentally induced problem in speech production (a bite-block speech condition) were compared between children with DAS and NS children.

Experiment 1: Durational pattern

In studies on normal durational patterns, various phonetic context effects have been found on segment duration. As was stated by Glasson (1984, p.87): “For each unit, duration is dependent on the factors of rate, intrinsic properties of the segment, stress, linguistic function, and phonetic context”. Harris (1984) found that vowels and consonants are coproduced by which means neighboring segments overlap (as also mentioned by Fowler, 1980). Previous studies had already shown that the duration of a vowel shortens as increasing numbers of consonants are added (Lehiste, 1976). Furthermore, forward shortening is reported as well, that is, a vowel shortens as increasing numbers of consonants precede it (Harris, 1984). These contextual influences of both consonants and vowels have been found in various studies in both adults and children (e.g., DiSimoni, 1974, in three-, six- and nine-year-old children). Smith (1978), comparing adult speech to child speech (a group of two-and-a-half to three-year-old and a group of four to four-and-a-half year-old children), found that although children revealed consistently longer word and segment durations than adults, the intrinsic properties (the proportional increments of segments) were similar in magnitude. Thus, the durational pattern of NS children shows particular intrinsic phoneme durations as well as adaptations to the context. For the utterances used in the present study we expected that fricative consonants would have longer durations than plosives and high vowels would be shorter than low vowels. Furthermore, segment durations would be adapted to the surrounding phonemes so that utterances with different phonemes have similar durations (e.g., vowels will be shorter in a fricative context than in a plosive context). These are all frequently mentioned mechanisms in phonetic research (Treiman, Straub, & Lavery, 1994).

In this experiment, correctly produced utterances of children with DAS were compared to those of NS children. Correct utterances were used to be able to make the comparison with NS children. Furthermore, when an error occurs in the speech of the children with DAS it is hard to tell what is the origin of this error. If the durational pattern of children with DAS is similar to that of NS children, despite overall longer durations, this indicates intact durational control processes involved. The longer durations are then likely to result from a slowing down of the process of speech production. (This might be interpreted as a compensatory strategy to enhance intelligibility by decreasing the number of production errors, as suggested by Darley, Aronson, & Brown, 1975.) An aberrant durational pattern, however, might reflect poor durational control. That is, systematic differences in durational patterns could be induced by some speech movements slowed down more than others, without being controlled by durational control processes. Whether these longer durations with possible aberrant durational patterns are a direct, primary, effect of the speech disorder concerning durational control or the effect of a compensatory strategy was further investigated in Experiment 2.

Experiment 2: compensatory speech production

At the outset of the second experiment, the question was: Given the fact that the speech of children with DAS is slower than normal speech, are the differences in durational pattern as reported in the literature an effect of extended movement durations (i.e., a compensatory strategy), or the result of poor durational control (i.e., the disorder)? Research in which a problem in speech production is induced, such as artificial perturbations like a bite-block, may provide the answer to the question whether durational differences are based on movement differences, or are caused by deviant durational control.¹ In the second experiment this is further explored.

NS children will be hindered when speaking with a bite-block, since this is an unnatural speech condition, which will result in longer segment durations (as was also found in numerous studies, e.g. Smith, 1987; Towne, 1994), particularly in longer consonant durations. Smith (1987) interpreted the increase of (consonant) duration, as compared to the normal speech condition, to be a result of biomechanical factors, because speakers are required to move their lips and tongue greater distances without assistance of the jaw. Furthermore, we argue that, since the bite-block fixes the jaw in a relatively 'open' position, high vowels will be more affected than low vowels. NS children will, thus, show longer durations in the bite-block condition due to unnaturalness of the speech situation, which requires slowing down in order to produce intelligible speech. The durational *pattern* is expected to remain stable, because possible differences in movement durations can be compensated for and the interdependency of the segment durations is not affected.

Subsequently it is interesting to study how do children with DAS adapt the durational pattern to the bite-block condition? Although various studies investigated bite-block compensation in children with DAS, with varying results (see for an overview Nijland, Maassen, & Van der Meulen, in press), hardly any study reported durational data. Towne (1994) found that phonologically disordered children were equally capable of compensating the diadochokinetic rate for the presence of a bite-block as normally speaking children (i.e. slower diadochokinetic rates but no interaction of group with speech condition). However, a subgroup of children with a more neuromotor disorder showed limited compensatory abilities. The durational data of the present study might therefore provide valuable information.

If children with DAS show similar effects as NS children, then apparently they were able to adapt to the induced problem in speech production in a similar way, by slowing down the process of speech production. It is then assumed that these children will also adopt this compensatory strategy of slowing down in a *normal speech* condition, which led to the slow speech and longer durations. However, if the children with DAS show aberrant durational adaptations in the bite-block condition as compared to NS children, then we might conclude that children with DAS differ from NS children with respect to compensatory abilities. Deviant durational patterns in the bite-block condition then resulted from differences in movement durations that could not be compensated for. This would indicate poor durational control.

¹ Another method to induce a problem in speech production could be a task in which speech rate must be increased. However, experience with children with DAS showed that they have problems with increasing speech rate without increasing speech errors too much. Since the acoustic analyses were conducted on the correctly produced utterances the bite-block task was given preference to increasing speech rate.

EXPERIMENT 1: DURATIONAL PATTERN IN A NORMAL SPEECH CONDITION

The question in Experiment 1 was whether children with DAS show similar intrinsic durational properties and similar interdependency of segment duration to the phonetic context as compared to NS children. For this, we studied the durational patterns in a normal speech condition.

Method

Participants

Eleven children with DAS (eight boys and three girls) and six normally speaking (NS) children (all boys) participated in this experiment. All children were native speakers of Dutch. They had been selected from a group of 19 children with DAS (14 boys and 5 girls; age range: 4;11 – 6;10 years) and 19 NS children (matched for sex, age, and dialect). (The acoustic analyses are rather time-consuming and therefore not all 38 children could be analyzed. No special criteria were adopted to select the eleven children with DAS and the six NS children from the larger group, although it was warranted that the children who were selected represented the age and severity range of the total group.) The children with DAS, pupils of special schools for children with speech and language disorders, met the clinical criteria described by Hall, Jordan and Robin (1993) and Thoonen, Maassen, Wit, Gabreëls and Schreuder (1996). This selection was based on samples of spontaneous speech, repetitive imitations of (meaningful and nonsense) words and brief phrases, and a diadochokinetic task. The selection criteria consisted of a complete phoneme repertoire with many phonemic errors, high frequency of consonant substitutions (and omissions in clusters), sequencing difficulties and groping behavior, inconsistent error patterns in repetitions of words and phrases, and difficulty in producing complex phonemic sequences (Hall et al., 1993). The selected children with DAS met all these criteria (see Table 1). Additional criteria (which the NS children also had to meet) were: no hearing problems, no language comprehension problems, no organic disorders in the orofacial area, no gross motor disturbances or dysarthria, and at least average nonverbal intelligence (Thoonen, Maassen, Wit, Gabreëls, & Schreuder, 1996).

Speech material

The speech material consisted of two-word utterances with simple CV-syllables, in which the relevant part was [dəCV] and C=/b,d,x,s/ and V=/a,i,u/. These bisyllabic nonsense utterances were spoken within the carrier phrase 'hé ... weer' (/he ... wɪ:r/) "hey ... again". This resulted in utterances like 'hé de ba weer', 'hé de di weer' ("hey the ba again", "hey the di again"). The participants were asked to repeat the utterances after the experimenter. Each utterance-type was elicited six times; the sequence of utterance-types was randomized. In case a child was not able to correctly produce an utterance (which was judged by the experimenter during recording), a second attempt was made straight away. This second production was subsequently used in the acoustic analysis. This resulted in 72 utterances (i.e., 4 consonants * 3 vowels * 6 repetitions) per participant.

An unidirectional dynamic microphone (Shure SM10A) mounted on a headset and a tape-recorder (Kenwood KX54) were used to record the speech samples. The headset kept the

microphone at a constant distance of 5 centimeters in front of the right corner of the subject's mouth.

Table 1: Individual scores on the selection tasks (percentage consonant substitution in meaningful and nonsense word-imitation task and mean number of syllables per second in Maximum Repetition Rate task) of the children with DAS and the normally speaking (NS) children on which the subject selection was based.

DAS	Age	Meaningful		Nonsense		MRR		Bite-block
		Subst	Substpl	Subst	Substpl	Mono-syll	Tri-syll	
#1	5;0	26%	76%	47%	68%	3.53	1.93	No
#2	5;1	24%	56%	64%	79%	4.12	3.38	No
#12	5;5	21%	79%	44%	76%	3.02	Unable	Yes
#13	5;6	22%	71%	38%	68%	3.64	2.21	Yes
#14	5;7	15%	60%	15%	30%	4.63	3.68	No
#17	5;10	17%	64%	67%	70%	4.63	Unable	No
#20	5;11	30%	60%	48%	72%	3.31	Unable	No
#21	5;11	31%	85%	-	-	3.20	Unable	Yes
#26	6;6	35%	78%	47%	65%	3.76	Unable	Yes
#28	6;10	40%	60%	39%	96%	4.23	3.61	Yes
#29	6;10	14%	56%	15%	60%	3.98	Unable	No
NS	Age	Subst	Substpl	Subst	Substpl	Mono-syll	Tri-syll	Bite-block
#54	4;9	7%	0%	12%	38%	4.29	3.52	No
#36	5;0	0%	0%	17%	64%	4.85	3.68	Yes
#42	5;3	3%	0%	6%	25%	4.74	4.05	Yes
#53	5;6	8%	0%	8%	20%	4.49	5.03	Yes
#58	5;6	5%	33%	9%	33%	4.44	5.08	Yes
#49	5;11	1%	0%	13%	0%	4.97	3.06	Yes

Note. Subst: percentage substitution of singleton consonants on syllable initial position. Substpl: percentage substitution of place relative to the number of substitutions (Subst). Mono-syll: mean number of syllables per second of monosyllable utterances (/pa/, /ta/, /ka/). Tri-syll: MRR in a tri-syllable utterance /pataka/.

Acoustic analyses

Speech samples that were phonemically correct (72 utterances per participant) were digitized at 25 kHz and the relevant sections [əCV] were spliced out, using the Kay Elemetrics Computerized Speech Lab (CSL) analysis system. Information of the oscillogram, the FFT-spectrogram, and the energy-window was used to determine the onset and offset of each segment, and markers were set at the segments' onsets and offsets. Vowel onset was appointed at the onset of the quasi-periodic waveform (determined from information of the oscillogram, harmonic information of the spectrogram, and increase in amplitude from the energy-contour); vowel offset was set at the end of the quasi-periodic waveform at the point at which a change in harmonics (in the spectrogram) and amplitude was observed (which also constituted the beginning of the consonant). These markers were used to determine segment duration. Reliability measurements on these analysis procedures, as described in (Nijland et al., 2002), showed a significant reliability

between three observers who independently placed markers in 72 utterances of two children (correlations between 0.78 to 0.99 over all markers). Second syllable durations were calculated by addition of the duration of the consonant with the duration of the second vowel.

Statistical analyses

The statistical significance of differences in segment durations between the two groups of children was determined using *t*-tests. Before this, the averaged values of the repetitions were calculated, in order not to violate the normality and homogeneity condition. Henceforth, significance levels were corrected for multiple comparisons using Bonferroni's correction and effect size was determined with ω^2 (Winer, Brown, & Michels, 1991). Subsequently, the effects of consonant and vowel on the durations were tested using analyses of variance in each group separately. Finally, the variability in measured segment durations was determined by calculating the variance attributable to the between subject factor Speaker, and the within subject factors 'Type' (utterance –type), and 'Error' (the variance within speakers that is attributable to the repetition of the same utterance) in both groups separately. In order to determine significance of differences in variance between groups F-ratios were calculated by dividing variances (i.e., $F(\text{factor}) = \text{var. (factor)-group a} / \text{var. (factor)-group b}$; see also Nijland et al., 2002; Nijland et al., 2003). Note that this does not concern a standard analysis of variance in which between-subject variance is divided by within-subject variance.

Results and discussion

Phonetic context effects

Figure 1 shows the mean segment durations of the NS children and the children with DAS (also see Table 2).² Figure 1a shows the mean durations in each consonant utterance type, Figure 1b shows the mean durations in each vowel context. To test whether the influence of phonetic context is similar for the children with DAS as compared to the NS children, the effects of the consonant in the utterances (the within subject factor Consonant type) and of the vowel in the utterance (within subject factor Vowel type) on the segment durations in both groups were tested for significance using separate analyses of variance. In Table 3 the results of the analyses of variance on the main and interaction effects of Consonant type and Vowel type on the segment durations and separately on the duration of the second syllable are presented for each group separately.

Firstly, both figures show (as expected) that the durations of the segments and the second syllable were longer in the children with DAS than in the NS children, independent of utterance type (all t 's(198) > 5.6; $p < .001$).

² The durations of the schwa were displayed in negative direction in order to better display the duration of the second syllable (i.e. the sum of the consonant and vowel duration).

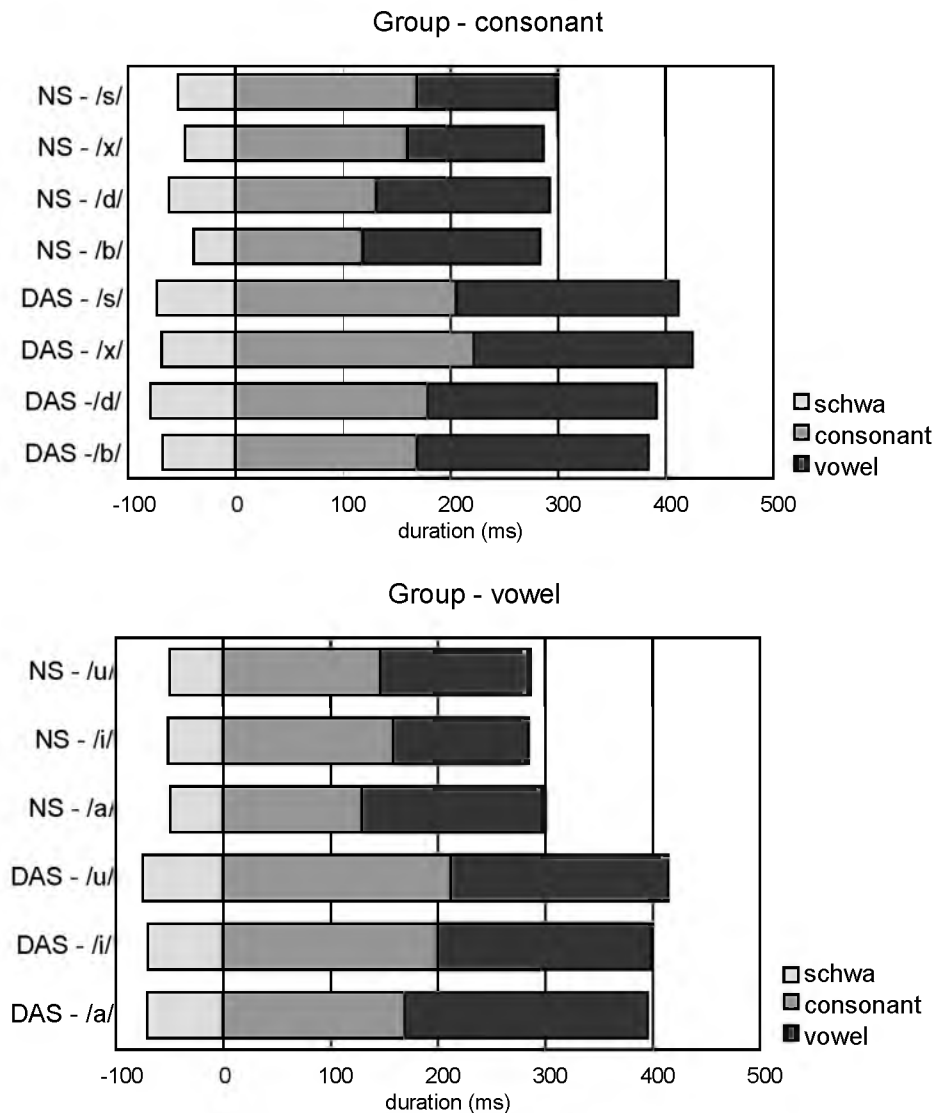


Figure 1. Mean durations of schwa, consonant, and vowel for each consonant type (upper figure, Figure 1a) and each vowel type (lower figure, Figure 1b) divided for the two groups: NS children and children with DAS.

Secondly, an effect of Consonant type is visible in Figure 1a in the NS children: the fricatives /s/ and /x/ were significantly longer than the plosives /b/ and /d/ (see Table 3, the factor Consonant type is significant on consonant duration). A similar effect of Consonant type was found in the vowel duration, although in the opposite direction, that is the vowels were significantly shorter when preceded by a fricative as compared to a plosive. This resulted in equal lengths of the second syllable in fricative and plosive utterances (see Table 3: the effect of Consonant type is significant on the consonant (intrinsic) duration, the vowel duration, and the second syllable duration in NS children). This suggests that the longer duration of the consonant was compensated by a shorter duration of the following vowel in normally children. In the children with DAS the effect of Consonant type on the durations tended to go in the same direction: fricatives were longer than plosives, which is compensated with a shorter duration of

the vowel. However, the effects were less clear and not significant in either the consonant or the vowel duration. The reason for not finding significant results in the durations of children with DAS might lie in larger variability, which is also visible in the higher coefficients of variance in Table 2 and will be discussed in more detail later.

Table 2: Mean segment and second syllable duration (in msec), with corresponding standard deviations (sd), and coefficient of variation (cv) in the normally speaking children (NS) and the children with DAS (DAS).

Group	Measure	Schwa	Consonant	Vowel	Second syllable
NS	N	72	72	72	72
	mean (ms)	50.2	144.5	145.2	289.7
	sd (ms)	14.4	32.5	32.8	36.5
	cv (%)	28.7	22.5	22.6	12.6
DAS	N	128	128	128	128
	mean (ms)	72.3	193.0	209.5	402.5
	sd (ms)	23.1	69.2	53.4	93.5
	cv (%)	31.9	35.9	25.5	23.2
DAS vs NS t (198)		7.3 ***	5.6 ***	9.3 ***	9.8 ***
ω^2		0.21	0.14	0.30	0.33

Note. *** $p < 0.001$; ω^2 indicates the effect size.

Table 3: F-values of analysis of phonetic context per speaker group. Presented are F-values from the analyses of variance on second syllable and segment durations. Factors are Consonant type (CONS) and Vowel type (VOW). For further explanation see text.

Group	Factor	Df	Schwa	Consonant	Vowel	Second syllable
NS	Cons	3,15	10.468 *** ¹⁾ (.677)	24.984 *** ²⁾ (.833)	24.277 *** ³⁾ (.829)	1.150 (.187)
	Vow	2,10	0.429 (.079)	58.787 *** ⁴⁾ (.922)	18.790 *** ⁵⁾ (.790)	2.152 (.301)
	Cons * Vow	6,30	1.915 (.277)	0.415 (.077)	1.346 (.212)	1.522 (.233)
DAS	Cons	3,29	2.135 (.181)	1.905 (.164)	1.299 (.118)	0.835 (.079)
	Vow	2,20	0.478 (.045)	11.543 *** ⁶⁾ (.534)	4.716 * (.320)	1.716 (.146)
	Cons * Vow	6,57	0.932 (.089)	1.485 (.135)	0.744 (.073)	1.202 (.112)

Note: significance, * $p < .05$, ** $p < .01$, *** $p < .001$; effect size (eta-square) is given between brackets. Post-hoc comparison: ¹⁾ $bx < sd$, ²⁾ $bd < xs$, ³⁾ $xs < bd$, ⁴⁾ $a < u < i$, ⁵⁾ $i < u < a$, ⁶⁾ $a < iu$.

Thirdly, another effect of Consonant type was found in the schwa of the NS children. The NS children produced significantly longer durations of the schwa in the utterances with /d/ and /s/ than in the utterances with /b/ and /x/. Presumably the carrier phrase is relevant in this. In the carrier phrase ([hed-əCV-wI:r]) the alveolar /d/ was uttered before the schwa, which led to a repetition of an alveolar articulation (either /d/ or /s/) instead of an alternation of movements as in ‘deba’ (tongue tip and lips) or the ‘dexa’ (tongue tip and tongue body). Since alternations of movements are faster than repetition of the same movement (Coover, 1923), the duration of the

schwa is longer in the repeating movement, that is in the /dədV/ and /dəsV/ -utterances. Again, the effect of Consonant type on the duration of the schwa seems to be operative in children with DAS (see Figure 1a), however, not significantly.

Finally, Figure 1b shows an effect of Vowel type in the utterances of the NS children, namely the vowel /a/ was longer than the vowel /u/, which in its turn was longer than the vowel /i/. The opposite effect is visible in the preceding consonant, in which the duration was shortest in the utterances with /a/, and longest in the utterances with /i/. As in the Consonant type effect, the longer duration of a segment seems to be compensated for by a shorter duration of the surrounding sounds; in this case the longer duration of the vowel is compensated in the preceding consonant. Both effects, intrinsic duration differences in the vowel and compensation, were significant (see Table 3), resulting in equal length of the second syllable in all utterances. For the children with DAS the effect of Vowel type was also significant in both vowel duration and preceding consonant duration: the low vowel [a] was longer than the high vowels [i] and [u], which was compensate for by shorter consonant duration before vowel [a] as compared to the high vowels [i] and [u]. Furthermore, no significant difference in second syllable duration was found due to Vowel type in the children with DAS.

Taken together, in the utterances of NS children clear intrinsic segment differences were found, namely fricatives were longer than plosives and low vowels were longer than high vowels. Furthermore, the segments showed compensatory effects in the surrounding sounds, that is longer consonants were followed by shorter vowels, and vice versa. In children with DAS, although very few significant effects were found, the effects tended to go in the same direction as in NS children. This means that the longer durations were not accompanied by significant deviant durational patterns.

Variability

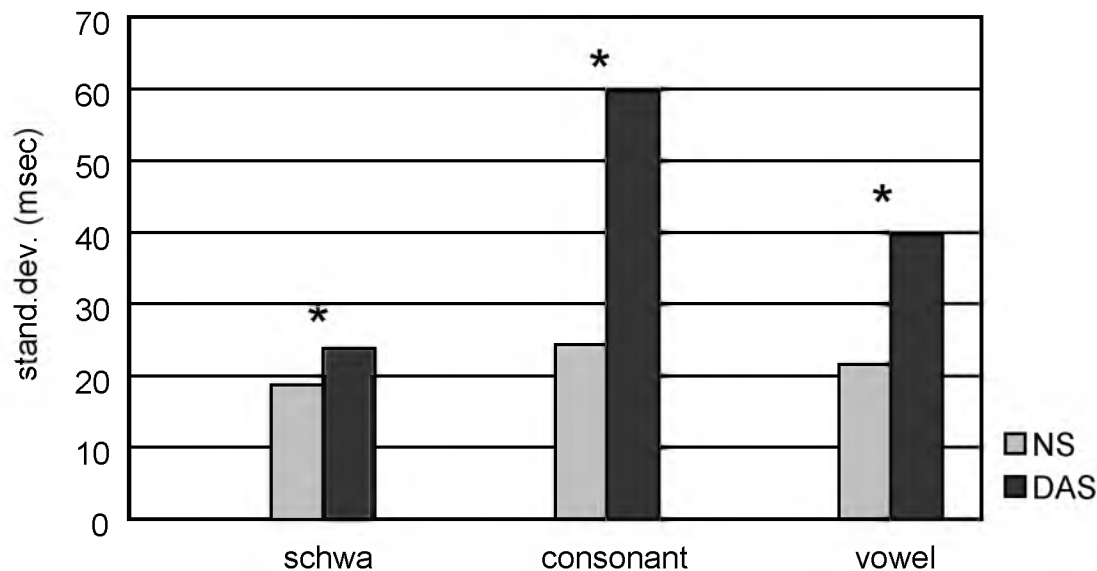
The standard deviations and the coefficients of variation of the segment durations, presented in Table 2, suggest that the children with DAS were more variable than the NS children. Previous studies indicated that within-speaker variability is higher in children with DAS than in NS children (Nijland et al., 2002; Nijland et al., 2003). In the present study we also analyzed variances of segment durations with respect within-speaker variability (repetition of the same utterances), between-speaker variability, and utterance type.

Strong effects of group were found in within-speaker variances. Figure 2 shows within-speaker standard deviations (square root of the variance) for both groups. Significant differences between groups are indicated with an asterisk in the figures, which shows that the children with DAS displayed overall larger within-speaker variability than NS children in all segment durations (schwa: $F(482,337)=1.62$, $\omega^2=0.27$; consonant: $F(484,339)=6.01$, $\omega^2=0.75$; vowel: $F(484,339)=3.40$, $\omega^2=0.58$; $p<0.05$).

The variances attributed to speaker variability and utterance type were also calculated. The only significant result was a higher between-speaker variability of vowel duration in the children with DAS as compared to NS children (vowel: $F(10,5)=5.49$, $\omega^2=0.74$, $p<0.05$) and a higher variability due to utterance type in NS children with regard to children with DAS in the vowel ($F(11,11)=2.91$, $\omega^2=0.48$, $p<0.05$). Furthermore, the variability in segment durations among

speakers and between utterance types did not differ significantly between NS children and children with DAS (Speaker - schwa: $F(10,5)=2.32$; consonant: $F(10,5)=2.99$; Utterance Type – schwa: $F(11,11)=1.75$; consonant: $F(11,11)=1.71$; ns).

Variability within speakers



Note. Significant effects are indicated with an asterisk.

Figure 2. Within-subject variability: standard deviations of segment duration in the utterances of the normally speaking children (NS) and the children with DAS.

To sum up, the results of Experiment 1 showed strong contextual interdependency in the segments of the NS children. That is, differences in intrinsic durations between plosives and fricatives, and between low vowels and high vowels, were both compensated for by the following vowel, and preceding consonant respectively. This combination of intrinsic durational differences and contextual interdependency was not found in the children with DAS, although the effects tended to go in the same direction as in the NS children. The absence of significant effects of Consonant type and Vowel type in the segment durations of the children with DAS might result from the larger variability that was found in the utterances of these children. However, apart from variability, the lack of durational control of speech also might be the reason for this lack of interdependency, which is also reported in studies of AOS (Ziegler & Von Cramon, 1985; 1986). Thus, the results of Experiment 1 were inconclusive, because the children with DAS did not show significantly different durational patterns as the NS children. Instead, the children with DAS showed the same tendencies as the NS children. Together with the high durational variability this leads to the conclusion that durational control in children with DAS is less strict. In order to further investigate durational control mechanisms in children with DAS, especially concerning compensatory strategies, the durational patterns were investigated in utterances in which speech production was complicated using a bite-block in the following experiment.

EXPERIMENT 2: DURATIONAL PATTERNS IN COMPENSATORY SPEECH PRODUCTION

In Experiment 1 slower speech rate and higher variability of duration were found in children with DAS. The lack of significant effects of intrinsic durational differences and contextual interdependency might reflect durational control problems in these children. It is not clear whether the slow speech and the deviant durational patterns reported in the literature are primary or compensatory symptoms, which will further be evaluated in this experiment. For this, we investigated whether children with DAS show effects similar to those experienced by NS children in a condition in which the presence of a bite-block induces a problem in speech production.

Method

Participants

The data of five children with DAS (out of 11 in Experiment 1) and five NS children (out of six in Experiment 1) will be presented here (see also the Appendix), since not all children were able to speak with a bite-block, partly because of lack of energy and concentration at the end of recording session including other speech tasks and partly because they were not able to clench the bite-block while speaking (also see Nijland, Maassen, & Van der Meulen, in press). These five children with DAS (two boys and three girls) were able to speak with a bite-block and also performed the same speech task at a second assessment 14 months later (the results of which will be presented in another paper).

Speech material and acoustic analyses

In this experiment the children produced the same speech stimuli as in Experiment 1, however, a bite-block was clenched between the first molars while producing the utterances. The size of the bite-block, made of two-component AV-putty standard (Silagum ®), was about 1 x 1 x 0.5 cm (h x w x d). A custom adjusted bite-block was made for each participant (Netsell, 1985). To prevent the bite-block from slipping during task performance, molar impressions were made by placing the bite-block between the participant's molars while it was solidifying. A spatula, 1 cm in height, clenched between the incisors, fixed the height of the bite-block. A string was pulled through the bite-block to prevent it from being swallowed. The analyses performed to obtain segment durations and second syllable durations were identical to the analyses described in Experiment 1.

Statistical analyses

As in Experiment 1, first the average values of repetitions were calculated. The main effect of bite-block on the durations was tested using Paired-Samples *t*-tests comparing the mean values (average of 6 repetitions) of the two speaking conditions (bite-block and normal speech condition). Bonferroni's correction was adopted on the significance level to correct for multiple comparisons (Winer et al., 1991). Analyses of variance were conducted in order to test interaction effects of the within subject factors: speaking condition (with or without bite-block) with Consonant type and speaking condition with Vowel type. This was done for each group

separately in order not to violate the assumption of homogeneity (Huck, Cormier, & Bounds jr., 1974). Analyses of variability in segment durations were similar to those in Experiment 1.

Results and Discussion

Figure 3 shows the mean durations of both groups in the normal speech and bite-block condition. Figure 3a shows the mean segment durations in each consonant context in both speech conditions; Figure 3b shows the mean segment durations in each vowel context in both speech conditions.³ Table 4 displays the mean durations (with corresponding standard deviations) of schwa, consonant and vowel in both speaking conditions of the NS children and the children with DAS.

Table 4: Mean segment durations and second syllable durations, with corresponding standard deviations (sd) and coefficients of variation (cv). Effect of bite-block is tested using Paired Samples t test.

Group	Condition	measure	Schwa	Consonant	vowel	second syllable
NS	-BB	mean	49.9	137.2	139.8	277.1
		sd	14.8	28.2	29.7	24.0
		cv (%)	29.6	20.6	21.2	8.6
	+BB	mean	42.4	165.8	166.5	331.3
		sd	20.0	30.6	33.7	37.5
		cv (%)	47.2	18.6	20.2	11.3
	-BB vs +BB	$t(59)$	2.65*	-10.62***	-7.94***	-12.47***
		ω^2	0.11	0.65	0.53	0.73
DAS	-BB	mean	75.1	210.0	235.0	445.1
		sd	30.2	93.2	58.8	114.4
		cv (%)	40.2	44.4	25.0	25.7
	+BB	mean	69.0	254.8	216.3	471.1
		sd	27.2	109.8	74.1	146.4
		cv (%)	39.4	43.1	34.2	31.1
	-BB vs +BB	$t(55)$	1.48	-3.16**	2.58*	-1.69
		ω^2	0.04	0.16	0.11	0.05

Note. Significance * $p < .05$ ** $p < .025$ *** $p < .01$; after Bonferroni's correction.

³ Note that these are the durational data of only five children with DAS, instead of 11 children with DAS in Experiment 1, are presented here.

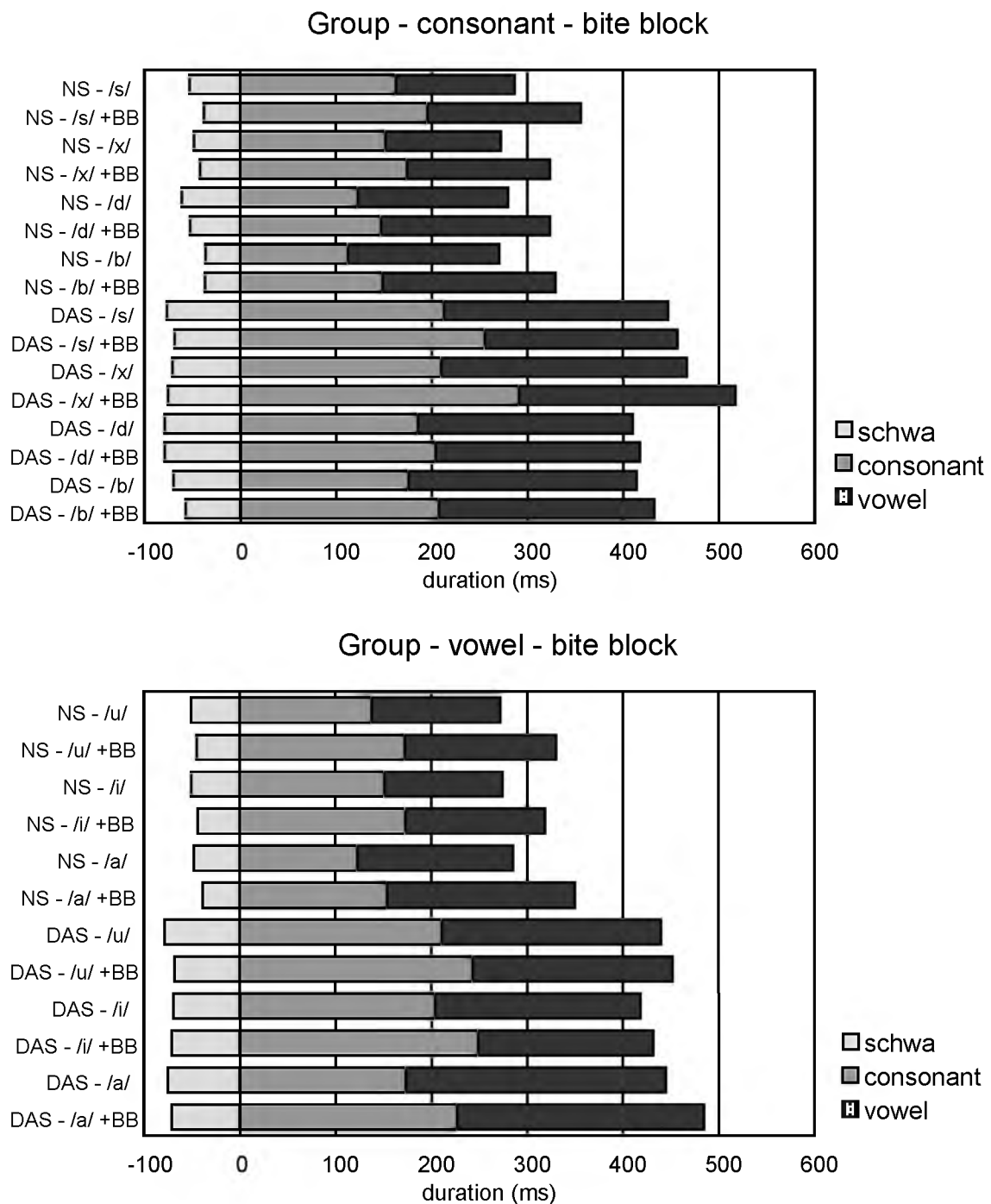


Figure 3. Mean durations of schwa, consonant, and vowel for each consonant type (upper Figure 3a) and each vowel type (lower figure, Figure 3b) divided for the two groups (NS and DAS) and two speaking conditions (normal speech and with bite-block).

Just as in Experiment 1, Experiment 2 shows that segment and second syllable durations were longer for the children with DAS than for the NS children, both in the normal speech condition as well as the bite-block condition (normal speech condition – schwa: $t(114)=6.08$,

$\omega^2=0.25$, consonant: $t(114)=5.77$, $\omega^2=0.23$, vowel: $t(114)=11.12$, $\omega^2=0.53$; bite-block condition – schwa: $t(109)=6.05$, $\omega^2=0.26$, consonant: $t(109)=6.06$, $\omega^2=0.26$, vowel: $t(109)=4.68$, $\omega^2=0.17$; $p<0.001$).

Furthermore, Figure 3a and 3b show that for NS children, speech production with a bite-block led to a significant increase of consonant duration and (second) vowel duration in all utterance types, and to a longer duration of the second syllable (also see Table 4). These results indicate that due to the bite-block the speech of the NS children was overall slower. As expected, the bite-block impeded the speech production, which resulted in slower speech.

Figure 3a and 3b and Table 4 show divergent results for the children with DAS with respect to the NS children. Like the NS children, the children with DAS also showed overall a significant increase of consonant duration due to the bite-block. In contrast to the NS children, the duration of the vowel did not increase, but showed a significant decrease due to the bite-block. As a consequence, the increase of consonant duration did not lead to a significant increase of the length of the second syllable. Thus, the children with DAS showed different effects of the bite-block manipulation on the consonant and vowel durations as compared to the NS children.

Another effect of bite-block was found in the schwa in the NS children: the schwa was shorter in the bite-block condition as compared to the normal speech condition. The pattern in Figure 3a suggests that the difference in schwa due to the following consonant as was found in the normal speech condition (see results of Experiment 1 in which the schwa was longer when it was followed by /d/ or /s/) was smaller in the bite-block condition. Such an effect was not found in the children with DAS in either one of the speech conditions.

Effects of Consonant type and Vowel type on the segment durations in bite-block speech are presented in Figure 3a and Figure 3b respectively. In order to study the role of the contextual influence on the durational pattern in bite-block speech, analyses of variance were conducted for each group separately. For this, the effect of Consonant type with Bite-block, and Vowel type with Bite-block was tested on the durations of the schwa, consonant, and vowel. Figures 3a and 3b (and the analyses of variance) clearly show that in the NS children bite-block speech led to an overall increase of the consonant and vowel durations independent of Consonant type (Figure 3a) or Vowel type (Figure 3b). This was corroborated with the findings of the analyses of variance. Effect on consonant duration: Bite-block $F(1,4)=46.4$, $\eta^2=0.92$, $p<.01$; Bite-block with Consonant: $F(3,88)=1.1$, ns.; Bite-block with Vowel: $F(2,88)=1.1$, ns. Effect on vowel duration: Bite-block $F(1,4)=27.1$, $p<.01$; $\eta^2=0.87$; Bite-block with Consonant $F(3,88)=0.99$, ns; Bite-block with Vowel $F(2,88)=1.1$, ns.. This means that plosives were equally influenced by the bite-block as fricatives and high vowels were equally influenced as low vowels.

Furthermore, Figures 3a and 3b show that the children with DAS tended to increase consonant durations due to the bite-block more in the fricative utterances as compared to the plosive utterances (Figure 3a), however not significantly (in consonant duration no significant effect of Bite-block $F(1,4)=6.2$; Bite-block with Consonant $F(3,81)=0.1$). Decrease of the vowel duration due to the bite-block was equal in both fricative and plosive utterances (no significant effect of Bite-block $F(1,4)=0.8$; Bite-block with Consonant $F(3,81)=0.9$). Furthermore, no significant effects of Vowel type with bite-block were found in children with DAS (no significant

effect of Bite-block with Vowel on consonant duration $F(2,81)=1.6$, nor on vowel duration $F(2,81)=1.4$).

Taken together, the NS children produced longer durations of consonant and vowel, and shorter schwa duration in the bite-block condition, independent of utterance type. Thus, speech production with a bite-block led to an overall increase of segment durations in NS children. The durational pattern, however, remained stable, which warrants intelligibility of the utterances. In contrast, the children with DAS showed an increase of segment duration due to the bite-block in the consonant, whereas the duration of the vowel decreased in the bite-block condition. No effect of bite-block was found in the duration of schwa, nor did the duration of the second syllable change. Thus, in these children the durational pattern changed due to the bite-block.

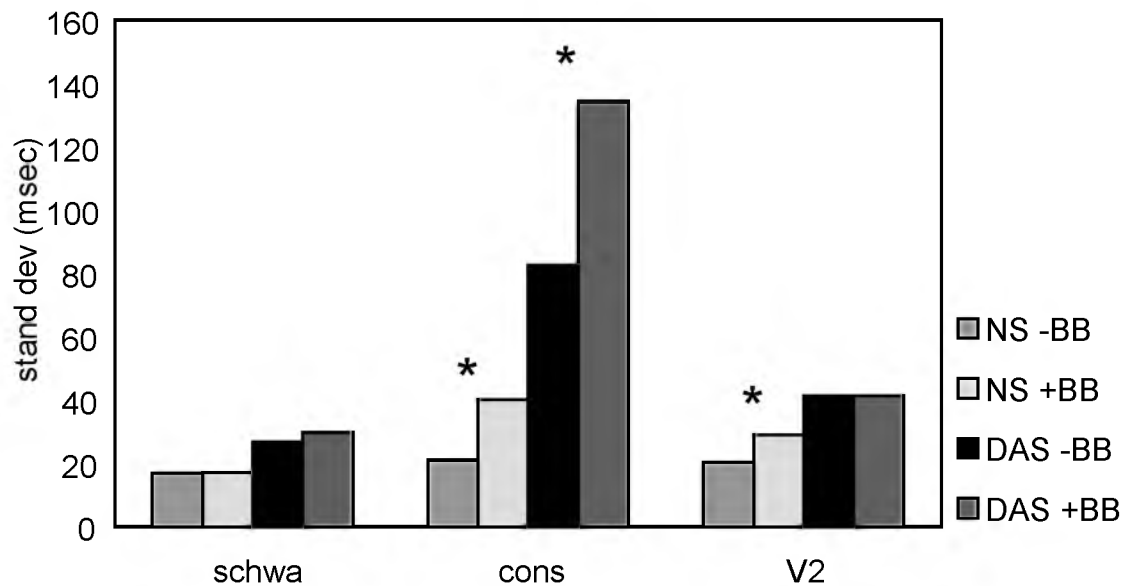
Variability

The standard deviations and the coefficients of variation of the segment durations that were presented in Table 4 suggested higher variability in the children with DAS as compared to the NS children. In Experiment 1 it was found that the within-speaker variability was higher in children with DAS than in NS children. In the Experiment 2 the effect of bite-block on variability is tested, in each group separately, and compared across groups. Most interesting is the effect of bite-block on the variability of utterance repetition (within-speaker variability), since it indicates the stability of the repeated production. Does the bite-block affect the stability of the utterance production? And, is this effect different in children with DAS as compared to NS children?

Strong effects of bite-block and group were found in the within-speaker variances. Figure 4 displays the within-speaker standard deviations for each group and speaking condition. An effect of bite-block was found in the within-speaker variability of the consonant and vowel durations. The within-speaker variability increased due to the bite-block in the consonant and vowel in the NS children (consonant: $F(286,212)=3.67$, $\omega^2=0.53$, vowel: $F(286,212)=2.01$, $\omega^2=0.30$, $p<0.05$; schwa: $F(286,198)=1.02$, ns.). In children with DAS due to the bite-block only the variability of the consonant duration increased (consonant: $F(189,183)=2.65$, $\omega^2=0.45$, $p<0.05$; schwa: $F(188,133)=1.23$, vowel: $F(189,183)=1.01$, ns.). Since the duration of the consonant also increased in the bite-block condition, the increase of variability as reported here might be merely an effect of correlation between mean and standard deviation. However, the correlations between the standard deviations and the mean durations were not overall very high to confirm this relation (correlation in DAS, schwa: 0.08, consonant: 0.55, vowel: 0.42; in normally speaking, schwa: 0.75, consonant: 0.26, vowel: -0.05).

Comparing both groups on within-speaker variability of segment durations, it was found that the children with DAS showed overall larger within-speaker variability than NS children; in all segment durations and in both speaking conditions (normal speech condition – schwa: $F(188,286)=2.55$, $\omega^2=0.38$, consonant: $F(189,286)=15.53$, $\omega^2=0.85$, vowel: $F(189,286)=4.09$, $\omega^2=0.55$; bite-block condition – schwa $F(131,198)=3.06$, $\omega^2=0.45$, $F(183,212)=11.23$, $\omega^2=0.83$, $F(183,212)=2.06$, $\omega^2=0.33$, $p<0.05$).

Variability within speakers



Note. Significant effects of bite-block are indicated with an asterisk.

Figure 4. Within-subject variability: standard deviations of segment duration in the utterances of the normally speaking children (NS) and the children with DAS, spoken in both normal speech condition (-BB) and bite-block condition (+BB).

As was mentioned at the beginning of this section, the variances were also calculated attributed to speaker variability and utterance type. However, both variances did not reveal any significant effect of bite-block in both groups. This means that the variability in segment durations among speakers and between utterance types did not change due to the bite-block.

To summarize, the NS children showed an increase of consonant duration as well as vowel duration in the bite-block condition. The durational pattern remained stable in order to produce intelligible speech. Children with DAS showed a different mechanism to adapt to the bite-block speech, in which consonant duration increased, but the duration of the vowel decreased, resulting in equal syllable durations but different durational patterns in both speech conditions. Thus, children with DAS differed in compensatory strategy due to the bite-block as compared to NS children. Furthermore, the variability results showed that in both groups the stability of the utterance was affected, that is, decreased due to the bite-block.

GENERAL DISCUSSION

In the present study, durational control of speech in children with developmental apraxia of speech (DAS) is studied in order to answer the question whether the slow speaking rate as found in DAS can be considered a result of the disorder or an effect of compensation. We argued that the durational pattern would reveal answers to the question whether the slow speech and deviant

durational patterns that are commonly reported in the literature of children with DAS are the result of a compensatory strategy in order to enhance speech intelligibility or a direct, primary, effect of the speech disorder. For this, two experiments were conducted, in which not only absolute segment durations were compared between children with DAS and normally speaking (NS) children, but moreover, the durational pattern was investigated. We acknowledge the fact that the small number of children might limit the statistical power of this study, therefore we included the effect sizes of the effects. However, within subjects effects were tested with regard to utterance type and speech condition in which each subject served at his/her own control. Furthermore, strong effects on the segment durations were found in the NS children, even though their number of subjects was limited with respect to the children with DAS.

The results of Experiment 1 showed clear durational patterns in the segment durations of NS children in which strong contextual interdependency of surrounding consonants and vowels was found. Differences in intrinsic durations between plosives and fricatives, and between low vowels and high vowels, were both compensated for in the duration of the following vowel and preceding consonant respectively. For instance, the longer duration of a fricative (as compared to a plosive) induced a shortening of the duration of the following vowel; and vice versa, a longer vowel /a/ (as compared to /i/ and /u/) was preceded by a shorter consonant duration. Although the durations of children with DAS tended to go in the same direction as in the NS children, this combination of significant intrinsic durational differences and strong contextual influences was not found. These results are in accordance with the results of other studies (e.g., Velleman & Strand, 1994). The absence of significant systematic effects, together with the high variability in duration that was found within the children with DAS, are interpreted as deficient durational control of speech in DAS (as was suggested before by Shriberg, Aram, & Kwiatkowski, 1997b; Velleman & Strand, 1994, and also in adult apraxia of speech by Seddoh et al., 1996; Ziegler & Von Cramon, 1985; 1986).

The results of Experiment 1 did, however, not answer the question whether the slow speech in DAS and the deviant durational patterns reported in the literature are primary or compensatory symptoms. This was further evaluated in Experiment 2, in which compensatory abilities to a bite-block manipulation in children with DAS were compared to those of NS children. Results showed that the NS children increased both consonant and vowel durations in order to adapt to the bite-block. This ensures us that the speech material (nonsense utterances) and speech condition (bite-block) enables us to study compensatory behavior in children. Thus, these children use a compensatory strategy by slowing down the process of speech production while controlling for the intrinsic durational differences and contextual interdependency by maintaining the durational pattern. This corroborated the results of Towne (1994). The children with DAS reacted differently to the bite-block as compared to NS children. Although, an increase of consonant duration was also found in these children, the duration of the vowel decreased. Apparently, these children do not use a similar compensatory strategy of equally slowing down as the NS children did. These aberrant durational patterns in children with DAS are assumed to result from differences in movement durations that are not controlled for. That is, some articulatory movements were affected more than others. Whereas the durations of the NS children give evidence of durational control in that the duration of both consonant and vowel

were increased due to the bite-block, the children with DAS do not show such a durational control. Apparently, durational control is not well developed in children with DAS.

Thus conceived, the deviant durational patterns that are commonly found in children with DAS, and were to some extent found in the present study, should be ascribed to a lack of durational control. That is, some movements are slowed down more than others without evidence of a 'higher' control mechanism. It is a leap of faith to suggest that, when children with DAS do not use durational control in a compensatory speech condition, this lack of durational control in a normal speech condition will neither be the result of compensatory effects. Yet, these conclusions corroborate the findings of former studies (Shriberg et al. 1997b, Velleman & Strand, 1994). Shriberg et al. (1997b) proposed that the problems that children with DAS exhibit in using stress (all syllables are stressed) result from a higher level of rhythm and prosody, rather than reflecting compensatory effects. Furthermore, a lack of durational control in the children with DAS can also explain the higher variability in duration of repeated utterances that was found in both speaking conditions in children with DAS as compared to NS children.

Although the present paper was not concerned with differential diagnosis, our results may enhance the knowledge on differential diagnosis when similar techniques are used as in the present paper and other speech disorders are compared. The long durations that are commonly found in other speech disorders might also exhibit deficient durational control, or be a result of slowed movement execution. In conclusion, the results of the present study, in which apart from quantitative differences (longer segment durations) qualitative differences (deviant durational patterns and different adaptation to experimentally induced problem) as well are found in children with DAS as compared to NS children, suggest that a lack of durational control is underlying the speech of children with DAS.

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Appendix: Table with individual data of segment durations.

Group	Child	Cons- type	Without bite-block									With bite-block								
			/a/			/i/			/u/			/a/			/i/			/u/		
			schwa	cons	vowel	schwa	cons	vowel	schwa	cons	vowel	Schwa	cons	vowel	schwa	cons	vowel	schwa	cons	vowel
DAS	1	/b/	48.3	109.0	157.8	71.0	116.8	172.2	46.0	151.5	159.3									
		/d/	76.5	96.0	187.3	83.0	129.5	208.5	93.4	125.0	205.2									
		/x/	53.2	167.8	138.4	73.4	195.8	170.4	83.0	195.7	155.5									
		/s/	50.8	187.0	162.5	69.3	191.0	170.5	67.6	195.0	156.0									
	2	/b/	68.7	153.2	189.3	68.6	177.0	205.4	62.8	208.5	208.7									
		/d/	52.2	168.0	162.6	91.3	223.0	181.5	67.2	174.5	171.7									
		/x/	75.8	178.3	221.2	78.4	172.2	196.6	92.5	245.3	194.7									
		/s/	87.4	190.8	201.4	74.6	200.0	169.4	75.4	196.2	192.0									
	12	/b/	49.7	98.0	234.8	77.5	100.8	161.0	57.7	147.3	159.5	29.6	98.0	219.8	64.3	113.0	138.0	47.4	140.9	155.8
		/d/	44.2	90.4	237.6	66.4	99.4	152.6	43.3	108.7	173.7	60.6	121.4	226.3	54.0	119.5	141.0	46.9	104.0	162.0
		/s/	48.0	152.5	212.0	58.8	188.0	152.3	58.3	166.3	156.0	42.0	161.8	210.0	42.2	228.6	123.5	38.2	183.7	136.7
	13	/b/	73.8	134.6	320.6	54.0	229.3	272.7	70.0	158.5	333.3	65.0	168.7	301.1	77.5	226.4	252.1	56.0	207.2	268.3
		/d/	94.7	130.5	289.5	83.3	166.0	267.0	79.0	200.5	291.3	97.3	188.4	275.2	83.3	195.5	221.3	63.0	180.8	229.1
		/x/	111.0	250.0	367.0	48.7	569.3	242.0	74.5	585.0	267.0	111.0	431.5	325.0	25.0	327.5	206.3	74.5	283.5	249.5
		/s/	79.3	194.8	291.3	85.6	251.6	234.6	87.4	287.8	267.8	67.1	233.4	282.6	85.6	288.1	196.6	87.4	317.0	207.8
	14	/b/	62.0	124.5	213.3	51.5	130.8	188.5	84.2	157.0	206.0									
		/d/	70.3	99.2	216.5	73.5	131.8	175.3	64.7	150.7	170.5									
		/x/	57.8	182.3	151.0	63.3	175.0	127.8	49.5	179.8	157.5									
		/s/	46.8	161.0	163.2	33.8	178.7	120.7	40.7	172.7	125.0									
	17	/b/	62.4	136.4	182.2	67.2	136.2	154.0	34.2	175.8	138.0									
		/d/	66.5	136.3	218.2	64.3	202.0	182.3	64.4	178.2	145.0									
		/x/	49.3	152.2	179.3	51.0	234.4	131.0	62.8	189.8	156.3									
		/s/	82.0	187.3	190.7	69.2	212.3	140.0	76.0	197.8	179.7									
	20	/b/	66.5	200.8	229.3	67.7	236.3	284.0	100.0	132.0	217.0									
		/d/	52.3	183.0	245.5	61.3	197.3	277.8	60.7	232.3	261.3									
		/x/	69.8	159.5	206.5	68.0	163.3	271.0	55.3	182.5	166.8									
		/s/	85.0	241.5	224.5	75.0	158.6	307.6	76.0	222.0	220.5									
	21	/b/	27.8	180.8	311.0	28.8	225.2	235.5	40.2	237.0	256.3	35.4	210.8	334.1	35.8	240.4	267.3	44.1	268.3	283.6
		/d/	57.3	221.3	269.7	56.8	262.8	213.8	55.5	277.2	227.7	83.7	225.0	318.8	62.6	324.7	224.9	67.0	295.8	272.6
		/x/	67.0	221.4	274.2	49.5	256.3	166.2	59.2	234.5	225.0	66.0	221.2	284.1	44.3	281.5	190.7	48.3	300.6	258.8
		/s/	88.2	324.8	265.8	51.7	264.3	202.3	49.0	271.0	237.4	89.3	326.2	309.8	71.2	330.9	202.4	70.2	281.9	292.8

	26	/b/	92.6	155.6	168.2	85.2	204.7	181.8	107.3	173.0
		/d/	106.6	176.4	163.2	120.0	202.7	164.0	181.7	397.7
		/x/				58.0	198.0	146.0	109.0	183.0
		/s/	112.7	146.0	184.3	54.0	155.0	195.0	156.0	158.0
	28	/b/	90.0	152.7	268.3	104.0	198.0	272.8	91.8	241.0
		/d/	98.3	203.8	290.8	87.2	192.0	205.8	64.7	249.0
		/x/	38.0	92.0	322.0	111.0	147.0	292.0	81.7	215.7
		/s/	80.3	167.5	314.5	70.6	226.0	294.6	68.5	242.0
	29	/b/	65.3	155.8	229.0	86.5	234.8	185.3	71.0	189.0
		/d/	116.6	156.0	257.8	102.3	154.3	181.5	114.2	187.5
		/x/	74.2	232.8	192.2	60.2	243.2	142.7	60.4	264.0
		/s/	86.8	211.8	215.0	77.6	235.2	189.8	94.6	239.4
NS	36	/b/	26.6	95.4	203.8	31.2	113.4	156.0	22.8	116.0
		/d/	72.0	103.3	206.7	57.6	146.8	128.6	73.0	122.8
		/x/	55.7	132.5	164.5	58.7	182.2	99.5	72.8	157.7
		/s/	51.7	160.8	163.5	65.7	174.7	91.0	61.8	168.5
	42	/b/	55.7	89.8	177.3	52.7	112.3	148.5	46.6	112.0
		/d/	80.4	97.4	199.2	55.4	137.0	151.4	62.4	134.2
		/x/	61.7	112.0	143.7	49.0	135.2	110.2	63.3	131.7
		/s/	60.2	130.8	130.5	53.7	148.0	90.8	59.3	131.8
	49	/b/	41.2	109.7	180.3	46.7	144.3	121.8	35.2	135.8
		/d/	55.3	99.5	166.5	64.3	117.2	142.5	65.8	98.3
		/x/	53.0	127.2	135.2	48.8	142.5	109.0	66.8	147.2
		/s/	55.5	146.2	154.7	71.0	165.7	122.8	56.8	152.0
	53	/b/	39.3	91.8	151.2	45.7	130.7	151.3	29.2	120.3
		/d/	54.5	118.3	168.0	58.3	158.2	153.3	69.7	110.0
		/x/	31.0	163.0	97.3	42.2	179.0	90.8	42.7	170.5
		/s/	44.8	182.8	142.7	46.3	199.5	108.2	40.2	174.8
	54	/b/	53.4	148.4	220.0	39.8	157.5	176.5	57.8	137.8
		/d/	64.4	162.0	216.2	72.2	175.4	138.0	53.5	180.8
		/x/	39.2	177.8	182.8	40.2	221.6	129.0	37.2	211.0
		/s/	57.7	161.3	200.7	69.8	213.2	123.0	37.3	220.3
	58	/b/	20.6	89.2	186.6	30.0	115.2	130.8	24.0	110.8
		/d/	53.2	112.2	174.3	58.7	150.8	132.5	40.7	131.7
		/x/	29.5	141.0	146.7	25.5	182.3	95.0	30.2	165.7
		/s/	24.2	146.2	156.0	54.5	190.8	127.5	48.7	167.8

163.0	82.8	218.0	164.2	76.7	207.1	160.7	96.7	215.5	136.9
152.7	108.8	172.6	163.9	109.0	230.8	147.5	119.9	213.0	160.3
169.0	98.8	188.8	179.8	79.7	194.1	137.3	114.3	193.3	153.8
163.5	97.3	174.4	156.5	82.8	177.1	158.8	132.0	197.4	156.3
234.5	79.3	159.8	276.0	90.1	186.8	258.9	75.2	216.3	251.3
237.3	85.9	147.7	297.0	69.8	196.8	208.9	83.0	206.8	228.0
275.0	51.5	138.3	323.3	101.5	189.0	279.8	70.6	256.1	237.8
336.0	64.6	171.1	278.3	68.4	222.0	247.6	64.3	221.6	303.3
221.0									
222.8									
172.6									
186.2									
176.3	31.2	109.9	216.9	38.8	137.5	157.3	28.1	141.3	174.1
138.4	58.5	115.9	234.1	33.3	151.4	135.3	46.0	143.7	152.2
140.2	42.9	142.7	185.8	43.9	196.2	105.8	53.3	168.0	124.7
127.8	31.3	171.9	186.5	40.3	186.1	92.8	50.9	185.1	127.3
145.8	51.7	96.2	186.5	52.0	122.2	165.8	51.8	138.6	154.2
140.8	56.0	107.4	196.8	61.5	137.5	141.5	80.2	159.2	146.7
106.2	59.5	134.2	132.3	62.7	144.9	135.6	63.9	134.0	122.0
86.2	64.9	149.4	163.3	58.5	156.0	118.8	64.0	153.3	118.6
134.2	32.9	119.8	188.8	43.9	141.8	140.8	27.8	143.3	142.0
138.7	45.7	112.9	184.8	60.0	123.7	148.3	58.9	107.8	147.6
123.7	45.9	134.5	140.5	42.0	151.9	118.0	53.3	152.4	134.1
123.2	40.1	159.0	162.3	47.3	191.3	113.1	40.0	167.2	141.1
119.5	33.8	121.0	172.8	45.8	156.1	157.2	33.6	146.1	143.4
147.0	68.4	141.1	205.4	80.5	153.4	161.3	78.8	131.5	157.1
108.5	37.8	171.8	142.5	45.0	190.8	108.0	44.3	184.6	129.3
86.7	45.7	191.8	164.5	51.6	202.6	123.1	37.6	196.5	123.3
203.2									
190.7									
145.2									
138.2									
179.6	28.0	107.4	187.5	27.0	138.8	155.4	23.9	139.3	191.4
167.8	43.8	140.6	186.5	40.8	159.4	134.3	43.1	134.8	167.7
142.0	24.7	155.4	163.4	31.7	194.8	135.1	32.1	183.9	147.1
147.7	25.3	181.5	180.4	41.5	204.9	140.1	44.0	190.4	172.4

CHAPTER SIX

Cognitive functions

Nijland, L., Maassen, B. (2002). Cognitive functions in Developmental Apraxia of Speech.
Manuscript submitted for publication.

ABSTRACT

Children are diagnosed as having developmental apraxia of speech (DAS) on the basis of specific speech characteristics, in the absence of problems in hearing, intelligence, and language comprehension. This does not preclude the possibility that children with DAS might demonstrate additional problems in other cognitive functions. In the present study we investigated four cognitive functions: motor functioning, memory, sensory functioning, and attention. The scores of 17 children with DAS at two measuring moments within 15 months, were compared with those of 17 normally speaking children. Results indicated a deviant development in sequential abilities. Furthermore, the scores on the sequential tasks (both sequential memory and motor functioning) were significantly correlated with severity of speech involvement. Additionally, the children with DAS had overall lower scores, but improved similarly on the second measuring moment when compared to normally speaking children, indicating a delay in development of about 15 months.

INTRODUCTION

Developmental apraxia of speech (DAS) is a neurologically based speech disorder that can be understood as a deficit in planning and regulating motor actions of voluntary and complex sequential speech movements. Its defining characteristics are: unintelligible speech due to a large number of consonant errors (especially substitutions and omissions), inconsistency of speech errors, articulatory abnormalities like groping behavior, and abnormal prosody (see e.g., Davis, Jakielski, & Marquardt, 1998; Hall, Jordan, & Robin, 1993; McCabe, Rosenthal, & McLeod, 1998). Furthermore, a set of exclusion criteria applies in order to exclude other underlying deficits that could explain the speech characteristics, such as dysarthria, mental retardation, hearing loss, receptive language disorder, and organic disorders in the orofacial area. Diagnostic discussions concentrate on differentiating DAS from phonological disorder, auditory processing disorder, and articulation deficits in children with SLI. Recent studies focus on the underlying deficits in children with DAS. Because speech normally develops in interaction with other psycholinguistic and cognitive functions (Locke, 1994), and DAS is a developmental disorder, studies to reveal the underlying deficits cannot be restricted to speech motor functions only, but should be taken in a broader perspective. In the present study we investigated cognitive functions in children with DAS, hoping to get more indications with regard to the underlying deficit of the disorder. The result may, furthermore, contribute to the discussion whether DAS can be viewed as a separate entity with associated problems (comorbidity) or as a symptom complex arising from a diversity of underlying deficits (Shriberg, Aram, & Kwiatkowski, 1997).

In order to choose the cognitive functions that might be interesting to investigate in children with DAS, we will take information processes of speech production and perception as starting-point and link other cognitive functions to these processes. Figure 1 displays a model of speech processing (slightly adapted from the model of Levelt, 1989). In former studies, we investigated whether the underlying deficit of DAS might be located in the levels of speech production below phonological encoding, that is phonetic planning or motor programming (Nijland et al., 2002; Nijland et al., 2003; Nijland, Maassen, & Van der Meulen, in press). The results of these former studies showed that children with DAS are impaired in both phonetic planning and motor

programming. In the present study we will investigate whether other cognitive functions, which can be associated with these levels of speech processing (including speech perception and feedback (monitoring) mechanisms) may give evidence of comorbidity.

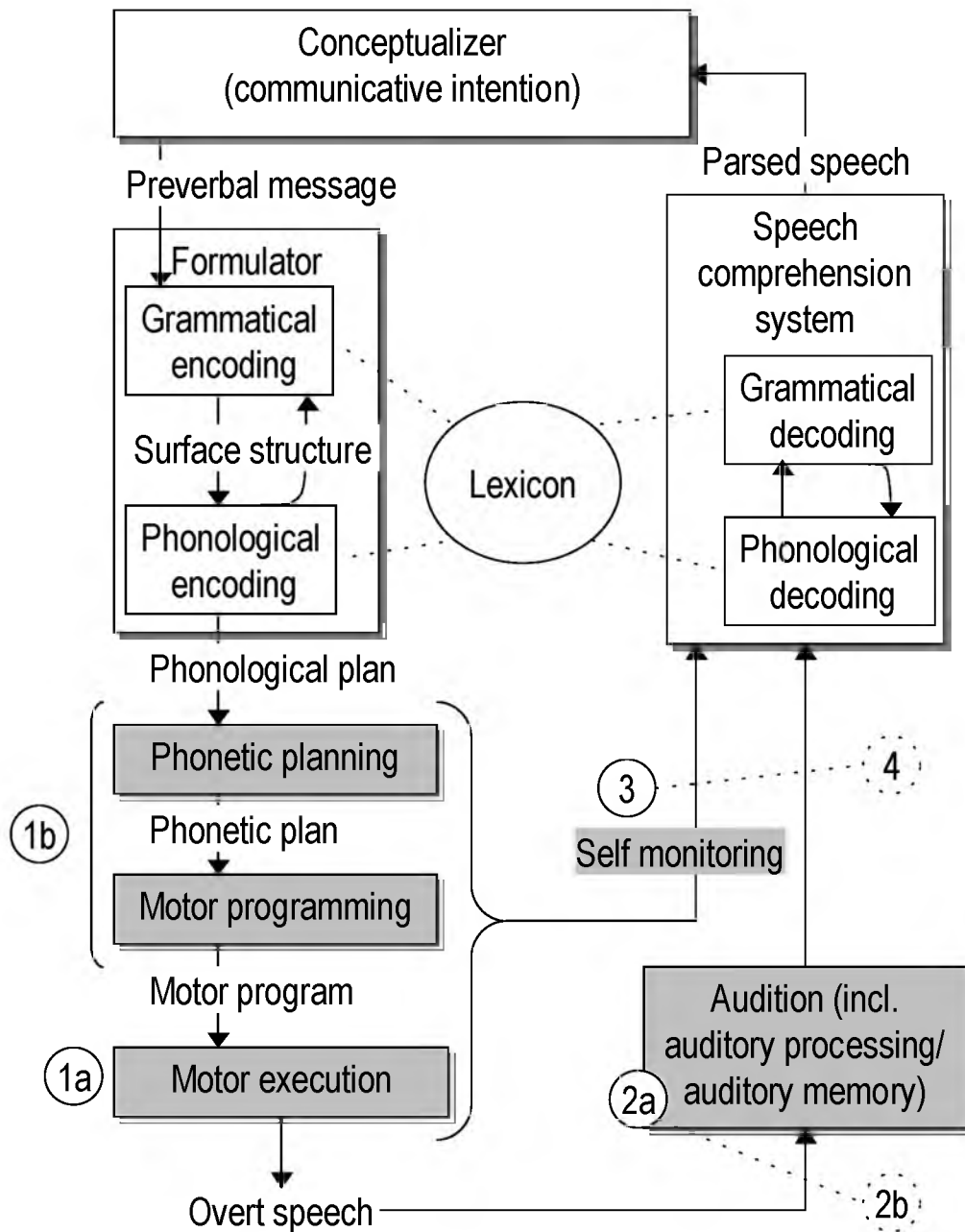


Figure 1. Model of speech processing, slightly adapted model of Levelt (1989).

We selected cognitive functions that can be associated with the processing stages that are crucial for DAS. The numbers 1a, 1b, 2a, and 3 in Figure 1 refer to these functions. The following four functions were studied in the present study: 1) motor functioning - (1a) motor

execution and (1b) motor planning, of which the latter is subdivided in psycho-motor planning (integration), sequential motor planning and auditory motor planning, 2) memory - (2a) auditory sequential and (2b) visual simultaneous memory, 3) sensory functioning, and 4) attention. The functions 'visual simultaneous memory' (2b) and 'attention' (4) could not directly be associated with processes in the model, but are linked with 'auditory sequential memory' and 'sensory functioning' respectively (displayed with a dotted line in Figure 1). These functions served as control functions and are assumed not to be affected in DAS. Before discussing the design and results of the present study, we will first give an overview of neuropsychological studies of DAS.

Cognitive functions in DAS

Although the disorder DAS is defined by its speech characteristics, most children with DAS also show impairments in other linguistic and nonverbal functions (Davis et al., 1998; McCabe et al., 1998). Also, 'soft neurological' signs such as motor coordination deficits (clumsiness) and mild motor retardation have been mentioned in the literature (see e.g., Ferry, Hall, & Hicks, 1975; Hall, 2000; McCabe et al., 1998; Velleman & Strand, 1994). However, little is published about neuropsychological research concerning children with DAS. The few studies, when not dealing with speech and language characteristics, focussed on motor behavior and memory capacity, in particular sequential memory (Dewey, Roy, Square-Storer, & Hayden, 1988). Problems in orosensory feedback, for example, have been mentioned occasionally as symptom of DAS, but were not thoroughly investigated (McCabe et al., 1998). Furthermore, studies on motor behavior and memory capacities in DAS show divergent results and interpretations.

Whereas some studies suggested that the difficulties in programming sequences of movements in DAS are restricted to the articulators (verbal and oral tasks, e.g., Aram & Horwitz, 1983), other studies assumed a more generalized motor sequencing disorder in DAS (Bradford & Dodd, 1996; Dewey et al., 1988; Yoss & Darley, 1974). According to Dewey et al. (1988) children with DAS had trouble with transitions between different movements within one motor sequence (e.g., pulling a knob and then turning it around). The same movement repeated a few times (as in finger-tapping) did not cause any problem.

In line with these findings of Dewey et al., Bradford and Dodd (1996) found that in comparison with other speech-disordered and control children, children with DAS scored low on both fine motor tasks and sequential oral motor movements. Bradford and Dodd interpreted the results as a deficit at the level of integrating sensory information into a plan of action (which is also a common explanation used in explaining limb apraxia (De Renzi, Faglioni, & Sorgato, 1982)), and at the level of coordinating speed and dexterity of complex movements. A different result was found in the study of Williams and Bishop (1992) who showed that both simple and complex manual tasks were slower in children with speech disorders as compared to normally speaking children.

These diverse results may firstly be due to differences in tasks administered. Aram and Horwitz (1983) used construction tasks, whereas Dewey et al. (1988) and Bradford and Dodd (1996) tested transitions between sequences of movements. Secondly, studies differed with respect to the selection of children on parameters such as age and speech characteristics. For example, Aram and Horwitz (1983) studied children within a wide age-range (4;4 to 13;2 years of

age) whereas the age-ranges in Bradford and Dodd's (1996) and Dewey et al.'s (1988) studies were much smaller (3;2 – 6;7 yrs and 4;5 – 7;1 yrs, respectively). Dewey et al. (1988) showed that a second group of children that was investigated did show severe difficulty in speaking (without evidence of DAS, average age 5;5 years), but did not show a generalized motor impairment. They suggested that the speech problem of these children was probably related more to a language or phonological planning problem. In contrast, Bradford and Dodd (1996) found that children with other speech-language problems than DAS showed similar low scores on sequencing oral movements as on verbal movements.

Turning now to memory functions, several studies investigated memory capacity in language disorders. However, these studies are conducted on diverse populations of children with phonological disorder and specific language impairment; only few studies have been reported on memory capacities in DAS specifically. Also in these studies, as in motor behavior studies, divergent results were obtained. Dewey et al. (1988) showed that spatial memory and memory for sequences was poorer in their children with DAS. However, they assumed that these problems in memory were not related to the difficulties in motor sequencing. Their first argument was, that also children with other developmental speech/language disorders had memory deficits without exhibiting motor sequencing difficulties, demonstrating dissociation of memory dysfunction from intact motor sequencing. Second, Dewey et al. (1988) found, in the children with DAS they studied, that the generation of sequences from memory was not performed more poorly as compared to producing spontaneous sequences, thereby questioning the role of memory in poor sequencing. Raine and colleagues (Raine, Hulme, Chadderton, & Baily, 1991) proposed another suggestion about the relation between memory and sequencing. They interpreted the lower short-term memory capacity in speech-disordered children to be causally related to a low speech rate. Also, Hulme and Roodenrys (1995) suggested that the development of verbal short-term memory skills seems to be intimately related to the development of speech production (and speech perception) mechanisms. Gathercole and Baddeley (1990) and Couture and McCauley (2000) found poorer recall performance in children with phonological impairments, which these authors attributed to interactions between short-term memory processes and aspects of phonological (long-term) storage. Thus, short-term memory deficits have been reported in studies on speech disorders as well as language disorders, which questions a specific relation between short-term memory deficits and DAS.

Aims of the study

Summarizing the above, although other dysfunctions additional to the speech problems in DAS have been mentioned in the literature, the research results are rather limited, and if available, controversial. To our knowledge, possible deficits in processing sensory information (except auditory functions) for instance have hardly been studied in children with DAS. The few studies that were conducted report diverse and controversial results. Furthermore, attention, which is a commonly studied function in developmental disorders, has not been studied in children with DAS specifically. The controversy in the reported results is due to differences in tasks and selection criteria. Therefore, in the present study, we used stringent selection criteria to select children that are clear cases of DAS, without additional problems in hearing, language

comprehension, gross motor functioning and intelligence (see Methods). Furthermore, we attempted to include a larger variety of functions, which were tested in a group of children with DAS and compared to normally speaking children. As stated above the functions concern motor functioning (execution and planning), memory (sequential and simultaneous), sensory, and attention. It was our premise that the functions sequential memory, motor planning, and sensory are related with the speech problems in DAS. The remaining functions, that is, simultaneous memory, motor execution, and attention, are not expected to be involved in DAS.

In the present study we investigated, firstly, whether a group of children with DAS showed different (i.e., lower) scores as compared to normally speaking children on one or more of these cognitive functions. Secondly, by comparing the scores on two measuring moments, in a period of 15 months, we were able to investigate whether a possible difference was the result of a delay in development or a deviant development. It is assumed that a period of 15 months is enough to show a significant effect of development in the normally speaking children between the results of the two measuring moments, and, moreover, to show a possible difference in development between the two groups. Thirdly, the results of the study allow us to address the issue of comorbidity of cognitive functions with speech functions. For this, we determined the correlation between the cognitive functions and the severity of speech involvement. Finally, homogeneity of the groups, especially of the children with DAS, was studied on the basis of clustering of the individuals within the research groups. In the literature a diversity of concomitant symptoms are reported with DAS (Hall, 2000; McCabe et al., 1998). Investigation of the homogeneity of the groups provides information about the specificity of cognitive functioning with respect to the speech disorder DAS.

METHOD

Subjects

Two groups of subjects participated in this study, a group of children with DAS and a group of normally speaking children. In order to come to a thorough selection of the children with DAS the following procedure was conducted. First, a group of children was selected by speech therapists of special schools for children with speech and language disorders, who also filled in a form concerning characteristics of DAS in these children. Subsequently, recordings were made of these children and these were judged on intelligibility of the speech and the possible involvement of dysarthria. On the basis of these results and an articulation test composed by Thoonen, Maassen, Wit, Gabreëls, and Schreuder (1996) the criteria described in Hall, Jordan, and Robin (1993) and Thoonen et al. (1996) were applied in order to select the clear cases of DAS. Additional exclusion criteria were: hearing problems, problems with language comprehension, organic disorders in the orofacial area, gross motor disturbances, dysarthria and below normal intelligence. Of the 70 children in the age group of 4;6 to 6;6 years who were referred to us by speech therapists as having DAS, only 19 (14 boys, 5 girls) were selected as clear cases of DAS. Neuropsychological data were collected twice in a period of about one to one-and-a-half year of

17¹ children of these children with DAS. A second group consisted of 17 normally speaking children, matched for (average) age, sex, and dialect region (see Appendix A for descriptive data of both groups of children). The age of the children with DAS at the first measuring moment was between 4;11 yrs – 6;10 yrs (mean age is 5;8 yrs); the normally speaking children were slightly younger between 4;7 yrs – 6;6 yrs of age (mean age is 5;6 yrs). The age of the children with DAS at the second measuring moment (about 15 months later) was between 6;1 yrs – 8;3 yrs (mean age is 6;11 yrs); the normally speaking children were between the age of 6;3 yrs – 8;0 yrs (mean age is 7;1 yrs).

Test-materials

Ten subtests were selected in order to evaluate the four main functions (mentioned above): 1) motor functioning, 2) memory, 3) sensory functioning, and 4) attention. Five of the ten subtests are derived from standardized assessment batteries: the Kaufman Assessment Battery for Children (K-ABC – Kaufman & Kaufman, 1983) and the Revised Amsterdam Children's Intelligence Test (RAKIT), for which age norms are available. The other five subtests did not have normalized scores.

- | | |
|------------------------|--|
| 1. Motor functioning | |
| a. Motor execution | <i>Finger tapping</i> (no age norms below six years of age). |
| b. Motor planning | <div> <div>Psycho-motor planning</div> <div>Sequential motor planning</div> <div>Auditory motor planning</div> </div> <div> <div><i>Labyrinths</i> (RAKIT).</div> <div><i>Hand movements</i> (K-ABC).²</div> <div><i>Auditory rhythm</i> (no age norms).</div> </div> |
| 2. Memory | |
| a. Sequential memory | <div> <div><i>Number recall</i> (K-ABC).</div> <div><i>Word order</i> (K-ABC).</div> </div> |
| b. Simultaneous memory | <i>Spatial memory</i> (K-ABC). |
| 3. Sensory | <div> <div><i>Oral sensory</i> (no age norms).</div> <div><i>Finger localization</i> (no age norms).</div> </div> |
| 4. Attention | <i>Deutsche SchulKonzentrationstest</i> |

Furthermore, hand preference of each child was determined on the basis of the hand that was used for writing/drawing. A detailed description of each subtest is given in Appendix B.

Statistical analysis

Before the statistical analyses were performed the values of the left hand and right hand were transposed, with respect to hand-preference. This means that the values of the left hand were

¹ Neuropsychological data of two children on the second measuring moment were not complete and therefore excluded from further analyses.

² This task (repeating a sequence of hand movements from memory) could be considered a motor planning task as well as a sequential memory task. Initially, it was considered a sequential motor planning task.

transposed to preference-hand values in case of a left-handed child, or non-preference-hand values in case of a right-handed child, and vice versa for the right-hand values.

In order to test whether the two groups differed and whether this changed over time, analyses of variance were performed with measuring moment as within-subject factor and group as between-subject factor. The main effects of group and measuring moment were determined as well as the interaction effect of group with measuring moment. A multivariate analysis of variance was conducted to investigate the main functions, and subsequent univariate analyses of variance were conducted on the separate variables.

In order to evaluate whether children with DAS show a deviance in development, rather than just a delay, we compared the results of the second measuring moment of children with DAS with the results of the first measuring moment of the normally speaking children. For this, analyses of variance were performed on the non-(age)normalized scores.

The relation between variables was tested using factor analyses for each measuring moment separately (in order to answer the question which subtests are significantly related to each other). Principal component analyses were conducted, in which factors with eigenvalues of 1 or greater were retained. Additionally, varimax orthogonal rotation with Kaiser normalization were used to enhance interpretability.

Finally, the factor scores of the individual children were used to group the children in clusters of children that were closest related to each other. A cluster analysis based on a simple Euclidean distance algorithm was used for this purpose.

RESULTS

Children with DAS versus normally speaking children

Table 1 displays the mean scores of the first and second measuring moments of both groups of children. The table clearly shows that children with DAS have overall lower scores than the normally speaking children. Furthermore, overall the values at the second measuring moment were higher as compared to the first measuring moment, in both groups.

Before conducting parametric analyses, the assumption of normality was tested using Kolmogorov-Smirnov tests for each variable on the non-(age)normalized scores. The results of this test showed that the scores of all variables were normally distributed (all K-S Z-scores < 1.33, ns.), except for three variables that had a strong ceiling- or bottom-effect. A bottom effect was found in 'auditory rhythm – both hands' at the first measuring moment of children with DAS (K-S Z = 1.63, $p < 0.01$) and 'Attention: omissions' at the second measuring moment of normally speaking children (K-S Z = 1.43, $p < 0.05$); a ceiling effect was found 'finger localization: 1 finger with looking' at both measuring moments of normally speaking children (measuring moment 1: K-S Z = 1.38, $p < 0.05$; measuring moment 2: K-S Z = 1.52, $p < 0.05$). Therefore, the effects of measuring moment and group were tested on composite scores (totals) of auditory rhythm and finger localization 1 finger (with looking was added to without looking). These composite scores were normally distributed (all K-S Z-scores < 1.00, ns.). Subsequently, analyses of variance were used to test the significance of the effects of group and measuring moment and

the interaction of group with measuring moment. The results of the analyses of variance are displayed in Table 2.

Table 1. Test scores (mean and standard deviation) of the children with DAS and the normally speaking (NS) children at the two measuring moments.

Variables	Children with DAS				NS children			
	First Moment		Second moment		First Moment		Second moment	
	Mean	Sd	Mean	Sd	Mean	Sd	Mean	Sd
1. Motor functioning	a. motor execution							
Fingertapping – preference hand	23.9	4.7	25.2	3.7	27.9	3.5	31.4	3.2
Fingertapping – non-preference hand	21.0	3.6	24.2	5.2	25.5	2.8	27.8	4.4
b. Motor planning – psycho-motor planning								
Labyrinths: RS	50.9	11.1	68.5	10.6	54.7	10.0	73.7	7.2
Labyrinths: SS	13.6	5.7	17.4	6.2	17.2	5.7	19.9	4.3
- sequential motor planning								
Hand Movements: RS	6.7	2.2	8.8	3.3	10.5	2.4	12.4	3.5
Hand Movement: SS	8.4	1.8	8.8	3.2	12.6	2.1	11.6	2.7
- auditory motor planning								
Auditory Rhythm – Preference hand	1.6	1.6	2.7	3.1	7.5	2.4	7.5	2.7
Auditory Rhythm – Non-Preference hand	1.1	1.2	2.5	2.1	7.2	2.5	6.4	3.3
Auditory Rhythm – Both hands	20.5	1.1	1.8	1.6	5.4	2.9	8.2	5.0
2. Memory	a. sequential verbal memory							
Number Recall: RS	5.1	1.6	7.1	1.5	9.1	1.7	11.4	2.1
Number Recall: SS	6.4	2.4	7.4	2.2	12.2	1.9	13.0	2.4
Word Order: RS	6.1	1.4	6.8	1.9	10.0	2.9	11.5	3.6
Word Order: SS	7.9	1.3	6.5	1.8	11.9	2.0	10.8	3.0
b. simultaneous memory								
Spatial Memory: RS	5.3	3.5	10.2	3.8	9.9	3.2	12.4	2.7
Spatial Memory: SS	8.1	3.1	9.7	3.6	13.1	3.2	11.2	2.3
3. Sensory								
Oral sensory	5.5	1.6	5.8	1.6	6.7	1.0	6.8	1.0
Finger localization: 1 finger with looking	16.9	4.0	18.1	2.4	18.9	1.6	19.5	0.8
Finger localization: 1 finger without looking	8.4	4.3	13.6	3.9	15.1	2.4	17.1	2.3
Finger localization: 2 fingers without looking	2.1	2.0	6.8	3.1	9.1	3.7	11.1	3.5
4. Attention								
Attention: mean row time	12.1	3.6	8.1	2.2	8.9	2.9	6.4	1.5
Attention: omission	6.4	4.0	1.9	2.3	2.7	2.5	21.1	1.9

Note. ¹ and ² indicate no normal distribution of the data due to a ceiling effect (¹) or a bottom effect (²).

The results of the multivariate analyses showed significant effects of both group and measuring moment on all main functions, and one additional interaction effect of group with measuring moment on *simultaneous memory* (the variable spatial memory). The latter was due to a larger increase in the children with DAS at the second measuring moment as compared with the normally speaking children. A post-hoc analysis showed that the difference between the normally speaking children and the children with DAS was not significant in the second measuring moment ($t(32) = 1.88$; ns.). This indicates that the children with DAS caught up with the normally speaking children on simultaneous memory. The above shows that the scores of the children with DAS were overall lower than those of the normally speaking children, and both groups had higher scores at the second measuring moment.

Table 2. Results of the analyses of variance with between subject factor *Group* and within subject factor *Measuring moment*.

Function	Variable	Df	Group (G)		Moment (M)		G*M	
			F	Eta2	F	Eta2	F	Eta2
1a. Motor execution – multivariate		2,31	9.19***	0.37	13.47***	0.46	2.28	0.13
	Preference hand	1,32	18.95***	0.37	17.24***	0.35	3.83	0.11
	Non-preference hand	1,32	11.38***	0.26	14.23***	0.31	0.45	0.01
1b. Motor planning – multivariate		3,29	18.93***	0.66	39.00***	0.80	0.22	0.02
Psycho-motor	Labyrinths	1,31	2.55	0.08	108.96***	0.78	0.13	0.00
Sequential motor	Hand movements	1,31	18.09***	0.37	13.73***	0.31	0.08	0.00
Auditory motor	Auditory Rhythm total score	1,31	57.34***	0.65	6.30*	0.17	0.43	0.01
2a. Memory sequential – multivariate		2,31	31.08***	0.67	36.45***	0.70	0.40	0.03
	Number recall	1,32	59.84***	0.65	74.75***	0.70	0.33	0.01
	Word order	1,32	40.35***	0.56	3.64	0.10	0.60	0.02
2b. Memory simultaneous – Spatial memory		1,32	10.85**	0.25	60.50***	0.65	7.16*	0.18
3. Sensory functioning – multivariate		3,28	20.53***	0.69	8.77***	0.48	1.95	0.17
	Oral sensory	1,30	8.76**	0.23	1.69	0.05	0.67	0.02
	Finger localization – 1 finger	1,30	26.10***	0.47	18.28***	0.38	4.54*	0.13
	Finger localization – 2 fingers	1,30	51.19***	0.63	18.08***	0.38	3.32	0.10
4. Attention – multivariate		2,31	7.63**	0.33	26.62***	0.63	3.27	0.17
	Mean row time	1,32	10.27**	0.24	44.17***	0.58	2.34	0.07
	Omissions	1,32	8.25**	0.21	27.11***	0.46	6.14*	0.16

Note. Significance is indicated with * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$. Multivariate F ratios are generated from Wilks's Lambda.

When considering the results of the analyses of variance on the separate variables we can observe slight differences in the effect of group and measuring moment. First, Labyrinths showed a significant effect of measuring moment, but no significant effect of group. This means that the groups did not differ significantly on psycho-motor functioning.

Second, the sequential memory variable 'Word order' showed a significant effect of group, but not of measuring moment. Thus, although children with DAS scored lower than normally speaking children on sequential memory, no significant change was found in either group on the second measuring moment. A similar effect was found in oral sensory, which showed a significant effect of group, but not of measuring moment. Finger localization, on the other hand, showed significant effects of both group and measuring moment and a significant interaction effect of group with measuring moment on finger localization one finger (with and without looking). The latter was due to a larger increase in time in children with DAS as compared to normally speaking children. Post-hoc analysis showed that the groups still differed significantly at the second measuring moment ($t(32) = 3.42; p < 0.01$).

Finally, also the attention variable 'number of omissions' showed a significant interaction effect of measuring moment with group. Again the effect of measuring moment was larger (a larger improvement) in children with DAS as compared to normally speaking children. Post-hoc analysis showed that the groups were no longer significantly different at the second measuring moment ($t(32) = 1.04; ns.$).

To summarize the above, the children with DAS had significantly poorer results than the normally speaking children. Although overall the scores improved at the second measuring moment, the difference between the two groups continues to exist, except for the scores on spatial memory. Besides this, the scores of finger localization one finger and number of omissions, also showed a significant interaction effect of group with measuring moment, in which the children with DAS showed a larger improvement at the second measuring moment as compared to the normally speaking children. Thus, the children with DAS seemed to catch up with the normally speaking children on spatial memory, finger localization and attention, however not completely on the latter two.

DAS, deviant or delayed development

In order to evaluate whether the children with DAS exhibit a deviance development or a delay in development, the results of the second measuring moment of children with DAS were compared to the results of the first measuring moment of the normally speaking children. Results of the analysis of variance are shown in Table 3. Combining the results presented in Table 1 with the significance levels from Table 3, it was found that the children with DAS in comparison with younger normally speaking children obtained equal scores on some functions, namely motor execution, simultaneous memory, sensory functioning and attention, but still had lower scores on the functions motor planning and sequential memory. The results of the univariate analyses of variance on the subtests showed that motor planning was not uniform. The scores on labyrinths (psycho-motor planning) were significantly higher in children with DAS, whereas the scores on the subtest auditory rhythm (auditory motor planning) showed lower scores in children with DAS as compared to (younger) normally speaking children. Thus, the cognitive functions of children with DAS do not show a harmonic profile: some functions show a delay in development of about 15 months, others functions are even more delayed and are likely to be deviant.

Table 3: Results of the analysis of variance that was conducted to test the effect of group in comparison of the results of children with DAS on measuring moment 2 and normally speaking children on measuring moment 1.

Function	Dependent Variable	Df	F	Eta2
1a. Motor execution – multivariate		2,31	2.59	0.143
	Fingertapping: Preference hand	1,32	4.99	0.135
	Fingertapping: Non-preference hand	1,32	0.77	0.024
1b. Motor planning – multivariate		3,29	19.53***	0.669
	Labyrinths ^a	1,31	14.03***	0.312
	Hand Movements	1,31	2.91	0.086
	Auditory Rhythm – total	1,31	36.01***	0.537
2a. Memory sequential – multivariate		2,31	9.90***	0.391
	Number Recall	1,32	12.39***	0.279
	Word Order	1,32	14.90***	0.318
2b. Memory simultaneous	Spatial Memory	1,32	0.06	0.002
3. Sensory functioning – multivariate		3,30	2.23	0.183
	Oral sensory	1,32	3.53	0.099
	Fingerlocalization: 1 finger	1,32	2.29	0.067
	Fingerlocalization: 2 fingers without looking	1,32	3.87	0.108
4. Attention – multivariate		2,31	0.72	0.045
	Mean row time	1,32	0.87	0.026
	Omissions	1,32	0.98	0.030

Note. Significance is indicated with * $p < 0.05$, ** $p < 0.01$, *** $p < 0.001$. ^a The scores on Labyrinths are higher in children with DAS (measuring moment 2) as compared to normally speaking children (measuring moment 1).

Relations between variables and with the speech disorder

In order to test which functions were highly related to each other, first, (inter-item) product moment correlation coefficients were calculated. Subsequently, factor analyses were conducted for both measuring moments separately. This resulted in three factors on both measuring moments. Since the factor analyses were conducted on a rather small number of cases ($N=34$), we have to consider the results with some reservation.

At the first measuring moment, the overall solution accounted for 74.9% of the variance. Factor 1 accounted for 55.7% of the variance (eigenvalue = 7.24), Factor 2 accounted for 11.4% (eigenvalue = 1.49), and Factor 3 accounted for 7.8% (eigenvalue = 1.09). Table 4 shows the factor loadings after orthogonal (varimax) rotation³. Each variable loaded on at least one factor (factor loading $\geq .50$). Based on the variable's highest factor loadings (which are underlined in Table 4), factor 1 comprises the sequential memory tasks (note that handmovements was considered to be a motor planning as well as a sequential memory task), auditory rhythm and finger localization. The latter two tasks are also related to sequential memory, since for repetition

³ We also conducted oblique rotations, since we did not know beforehand whether the factors were correlated or not. This resulted in similar distributions of factor loadings, therefore the results were not displayed or discussed here.

of a rhythm or indication of the order of finger touching, storing information in and retrieving information from sequential memory is required. Factor 2 consists of psycho-motor planning, simultaneous memory, sensory functions and attention. Handmovements and finger localization 1 finger loaded on factor 1 as well as factor 2. Factor 3 contains motor execution.

Correlations were determined between the factor scores of the three factors and speech data obtained during the first recording session. Significant correlations were found of the first factor with MRR(=Maximum Repetition Rate)-monosyllabic ($r = 0.44, p < 0.02, n = 34$), with MRR-trisyllabic ($r = 0.66, p < 0.001$); and also with number of consonant substitution in meaningful utterances ($r = -0.76, p < 0.001$), number of substitution in place of articulation in meaningful utterances ($r = -0.80, p < 0.001$) and number of consonant substitutions in nonsense utterances ($r = -0.78, p < 0.001$), number of substitution in place of articulation in nonsense utterances ($r = -0.65, p < 0.001$). Factor 1 did not show significant correlations with number of substitutions of manner or voicing. All other factors did not significantly correlate with the speech data, except for factor 3 with MRR-trisyllabic ($r = 0.37, p < 0.05$).

Table 4: Factor scores of the variables on the first and second measuring moment.

Variables	Moment 1			Moment 2		
	I	II	III	I	II	III
Fingertapping – preference hand	0.22	0.17	<u>0.90</u>	<u>0.65</u>	0.23	<u>0.56</u>
Fingertapping – non-preference hand	0.27	0.30	<u>0.84</u>	0.19	0.42	<u>0.74</u>
Labyrinths	-0.05	<u>0.72</u>	0.48	0.08	<u>0.81</u>	0.23
Hand Movement	<u>0.68</u>	<u>0.55</u>	0.07	<u>0.63</u>	<u>0.51</u>	-0.08
Auditory Rhythm – total	<u>0.82</u>	0.32	0.31	<u>0.66</u>	0.29	0.01
Number Recall	<u>0.77</u>	0.27	0.39	<u>0.86</u>	0.22	-0.06
Word Order	<u>0.80</u>	0.06	0.29	<u>0.80</u>	0.25	0.18
Spatial Memory	0.46	<u>0.55</u>	0.49	0.30	<u>0.77</u>	0.21
Oral sensory	0.32	<u>0.59</u>	0.34	0.14	<u>0.74</u>	0.13
Finger localization: 1 finger	<u>0.62</u>	<u>0.60</u>	-0.01	0.43	<u>0.58</u>	0.37
Finger localization: 2 fingers without looking	<u>0.83</u>	0.20	0.08	<u>0.75</u>	0.02	0.21
Attention: mean row time	-0.23	<u>-0.69</u>	-0.37	-0.26	<u>-0.60</u>	-0.14
Attention: omissions	-0.29	<u>-0.70</u>	-0.06	0.03	-0.15	<u>-0.84</u>

Note: Extraction Method: Principal Component Analysis. Rotation Method: Varimax with Kaiser Normalization.

The factor analysis on the data of the second measuring moment also resulted in three factors. The overall solution accounted for 67.0% of the variance. Factor 1 accounted for 46.0% (eigenvalue = 5.98), Factor 2 accounted for 12.8% (eigenvalue = 1.67), and Factor 3 accounted for 8.2% (eigenvalue = 1.07). Looking at the factor loadings in Table 4, the factors of the second measuring moment show similar patterns as compared to the first measuring moment. This repetition of factor patterns increases the reliability of the results of the factor analyses.

Clusters of the children

The above results showed that the scores of several cognitive tasks, especially concerning sequential tasks, were significantly correlated with consonant substitution rates. It is, however, unclear whether the subjects in both groups showed homogeneous patterns in cognitive functions. In order to investigate the homogeneity of the groups, cluster analyses were conducted on the individual factor scores of the three factors. For the first measuring moment, this resulted in four significant clusters in which the subjects were subdivided (factor 1 $F(3,30) = 22.37$, factor 2 $F(3,30) = 19.01$, factor 3 $F(3,30) = 14.33$; $p < 0.001$). Cluster 1 comprised seven children with DAS and one normally speaking child, cluster 2 twelve normally speaking children, cluster 3 four children with DAS and one normally speaking child, and cluster 4 six children with DAS and three normally speaking children. To illustrate the subdivision of the children in clusters, the factor scores of Factor 1 and 2 are plotted Figure 2a, and the clusters are represented within the circles. Figure 2a clearly shows that most normally speaking children clustered within a small cluster. The children with DAS were more heterogeneous. All children with DAS showed lower factor scores on Factor 1 as compared to the normally speaking children (equal or lower than 0), except for one normally speaking child who also scored lower than zero on factor 1. Furthermore, the children with DAS showed large variability on Factor 2. This heterogeneity emerged in the cluster distribution of the children in that most normally speaking children clustered within one cluster, and the children with DAS were subdivided over three clusters.

The factor scores of the second measuring moment were also significantly clustered in four clusters (factor 1 $F(3,30) = 26.24$, factor 2 $F(3,30) = 20.46$, factor 3 $F(3,30) = 5.45$; $p < 0.01$). Cluster 1 included seven children with DAS and two normally speaking children, cluster 2 one child with DAS and twelve normally speaking children, cluster 3 includes five children with DAS and two normally speaking children, and cluster 4 includes four children with DAS and one normally speaking child. As illustration, Figure 2b shows a scatterplot of Factor 1 and 2 and the subdivision in the four cluster of the second measuring moment. Again, as in the first measuring moment, the children with DAS show more heterogeneity in factor scores and Factor 1 displays higher scores for almost all normally speaking children as compared to the children with DAS.

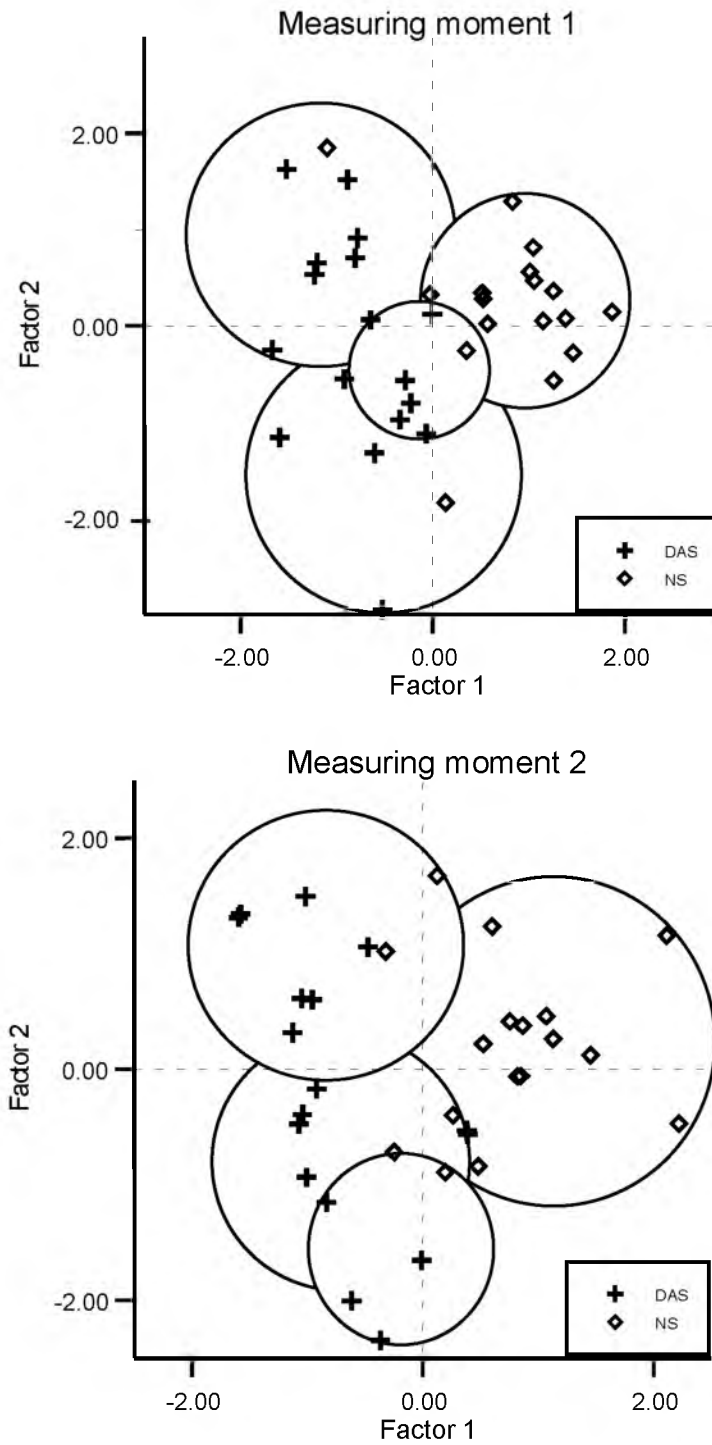


Figure 2: Scatterplot of individual factor scores on Factor 1 with Factor 2 on the first measuring moment (upper figure 2a) and second measuring moment (lower figure 2b). The individual children cluster within the circles

DISCUSSION

The aim of the present study was to investigate dysfunction in cognitive functions additional to the speech problems in children with DAS when compared to normally speaking children. Since speech and language develop in interaction with other cognitive functions, and developmental apraxia of speech concerns a developmental disorder (as indicated in the name), investigation of the cognitive functions in DAS seems necessary for our understanding of the disorder. Cognitive functions were related to successive levels of the speech production process that were investigated in our earlier studies (Nijland et al., 2002; Nijland et al., 2003; Nijland et al., in press). The present study addressed four questions, concerning 1) the comparison of cognitive functioning between children with DAS and normally speaking children, 2) the development of cognitive functioning with respect to an overall delay versus deviance, 3) the coherence of diverse cognitive functions and the comorbidity of these functions with severity of speech involvement, and 4) the homogeneity of comorbidity patterns of the individual children.

Firstly, in the comparison of the two groups the results showed that the children with DAS had overall poorer results than the normally speaking children. Although both groups show an improvement at the second measuring moment (overall higher scores), the scores of the children with DAS were overall lower than those of the normally speaking children, except for the simultaneous memory task, which was no longer significantly different at the second measuring moment. Besides this, also two other subtests (finger localization with one finger and number of omissions in the attention task) showed a larger improvement in children with DAS at the second measuring moment, however, not enough to completely catch up with the normally speaking children. These results suggest that children with DAS show an overall delay in development, which remains more or less stable during development.

Secondly, this suggestion of an overall delay versus a deviance was further investigated, by comparing the results of the second measuring moment of the children with DAS with those of the first measuring moment of the normally speaking children. The results of this comparison supported our assumption that the functions motor execution, psycho-motor planning, simultaneous memory, sensory functioning, and attention are not specifically related to DAS. This means that a possible delay at a certain stage in development is likely to make up arrears in a later stage. In contrast, other functions with regard to sequential memory (i.e., number recall, word order, and auditory rhythm) were assumed to be specifically related to DAS. As it appeared, children with DAS still performed poorer with respect to these functions, even when compared with normally speaking children who were more than a year younger and matched on the other functions.

The delay in development can be interpreted as a concomitant effect of the disorder of a rather general nature. Locke (1994) reviewed results of various studies that reported poorer performance of language delayed children on tasks that seemingly had nothing to do with speech. He suggested that the compensatory activity resulting from the deficit competes with other activities that consequently suffer from this competition. Such a mechanism seems to be at play here. In children with DAS the speech deficit competes with other functions so that this may consequently delay the development of the other functions. Thus, the results support the hypothesis that children with DAS are deviant on the functions concerning sequential abilities

(both sequential memory and sequential motor functioning), and overall delayed. The following paragraph discusses the possibility of a specific relation between these cognitive functions and severity of speech involvement.

Thirdly, the cohesion between diverse cognitive functions was investigated using factor analyses. A factor analysis of the first measuring moment resulted in three factors, which could be described as follows: factor 1 comprised Sequential memory functioning, factor 2 consisted of (psycho-)motor planning, simultaneous memory, sensory functioning and attention, and factor 3 contained motor execution. Furthermore, significant correlation coefficients were found between factor 1 and the speech data. This indicates a relation between low scores on the sequential memory tasks with the speech disorder DAS. Now the question arises whether the correlation between functions changes during development. The results of the factor analysis of the second measuring moment showed a similar pattern as compared with the first measuring moment. When combining the results of the factor analyses with the results discussed in the preceding paragraphs we can state that sequencing plays an important role in the deficit DAS. The children with DAS were more impaired on sequential tasks, even when compared with younger normally speaking children that were level-matched on other tasks, and furthermore, scores on sequential tasks were significantly correlated with severity of speech involvement. Thus conceived, problems in sequencing seems to be associated with the speech disorder DAS.

Finally, the homogeneity of the groups was investigated using cluster analyses. The cluster analyses showed that the group of children with DAS was more heterogeneous than the group of normally speaking children. Whereas the normally speaking children clustered more or less in one group, the children with DAS were split up in three groups. Figure 2a and 2b illustrated that the individual factor scores on factor 1 (the sequential tasks) were lower in all children with DAS as compared to normally speaking children. Although the children with DAS were heterogeneous with respect to the scores on most cognitive functions, on the sequential tasks they were homogeneously impaired. This further corroborates the finding that factor 1 is correlated with severity of speech involvement.

Concluding the above, the results of the different analyses all corroborate the suggestion that children with DAS as compared to normally speaking children are impaired on sequential tasks (both sequential motor and sequential memory tasks). All children with DAS had concomitant deficits on sequential tasks, but the profiles of the remaining functions were rather diverse. The results contribute to the discussion whether DAS can be viewed as a separate entity with associated problems (comorbidity) or as a symptom complex arising from a diversity of underlying deficits. There are two options here: 1) DAS is a symptom complex (among which the typical speech symptoms occur) resulting from an underlying general deficit in sequencing, or 2) DAS is a deficit in sequencing that is restricted to speech movements, with an underlying general deficit in sequencing in most children with DAS. As long as we are not certain whether all children with DAS show a general deficit in sequencing and whether all children with a general deficit in sequencing have DAS, the second option seems to be the safest. With this conclusion in mind, we will speculate on the neurological origin of the speech disorder DAS.

Neurological studies

Although DAS is known as a neurogenic speech disorder, in neurological studies evidence for brain lesions or abnormalities in children with DAS is sparse. However, ‘soft neurological’ signs such as motor coordination deficits (clumsiness) and mild motor retardation have been mentioned in the literature (see e.g., Ferry et al., 1975; Hall, 2000; McCabe et al., 1998; Velleman & Strand, 1994). In his search for focal or diffuse brain lesions in children with DAS, Horwitz (1984) concluded that neither consistent neurological findings nor a specific localizing anatomical basis for the clinical manifestations of DAS could be found. In contrast with acquired apraxia (Dronkers, 1996), developmental apraxia is not caused by brain damage but by a delayed or disordered maturation of the brain. The areas of the cortex that are important to praxis and that are impaired in acquired apraxia are the parietal and temporal cortex. Although, we do not know whether DAS might also be located in these areas, a lot of different problems, including problems with praxis of speech, can be the result of delayed or disordered maturation (these areas are also the last ones to mature in normal development). Delayed neurological development is also suggested in developmental language disorders, (Locke, 1994). In contrast to the above studies that located apraxia of speech in the parietal and temporal cortex, Roy (1978) related apraxia to a frontal lobe pathology, resulting in deficits in the ability to organize speech movement sequences and, moreover, general organization problems. The prefrontal cortex “comprises the medial, orbital (...) lateral and polar areas. These regions have rich connections with posterior association cortex, with the limbic system, parts of the corpus striatum (caudate and putamen), hypothalamus and mesencephalon. Given the wide pattern of connections with systems involved in sensory, mnemonic, motivational and motor processing it is hardly surprising that it has been impossible to define a narrow role of this region. However, broadly speaking the cognitive deficits following frontal damage frequently involve disruption of an action system, either through failure to initiate appropriate activity, or to integrate relevant information into the system guiding the current action.” (Crawford, Parker, & McKinlay, 1992, p. 269). There is ample evidence, both from animal and human studies, that the (pre)frontal cortex is the neuro-anatomical substrate of the ‘executive functions’ (Crawford et al., 1992; Denckla, 1996; Klenberg, Korkman, & Lahti-Nuuttila, 2001). The concept of ‘executive functions of the brain’ was introduced by Luria (1966) and Lezak (1982) (in Crawford et al., 1992). This term refers to cognitive abilities responsible for controlling and coordinating performance in complex cognitive tasks, such as planning, programming, regulation and verification of goal-directed behavior. Executive function is considered to be a cofactor with other processing deficits in the acquisition of basic skills, and is therefore important in new or unknown situations in which routine behavior is insufficient. Dysfunction of the executive functions emerge in problems in planning and organization.

More and more studies try to find relations between speech development disorders and abnormal brain structure. Recently, a follow-up study of a girl with developmental dyspraxia (not specifically of speech) showed an incompletely myelinated (immature) corpus callosum at the age of 5;6 years. Follow-up after two years showed no change in neuromotor assessment nor in speech production, which suggested that speech motor control is under the control of the same maturational constraints as motor development (Le Normand, Vaivre-Douret, Payan, & Cohen,

2000). In a recent study on the members of a family (the KE family) who are affected by a severe disorder of speech and language, abnormal development of the caudate nucleus were related to the impairments in oromotor control and articulation (Watkins et al., 2002). Also functional magnetic resonance imaging (fMRI) studies are searching for answers to the problems in apraxia of speech by measuring differences in brain activity (e.g., Rieckel et al., 2000). However, to date, evidence supporting that DAS is a 'neurogenic' disorder sticks to genetic aspects (DAS runs in the family and the preponderance in males) and the elevated incidence of comorbidities.

Results of our earlier studies showed that children with DAS exhibit problems in planning and programming of speech movement sequences (Nijland et al., 2002; Nijland et al., 2003; Nijland et al., in press). In addition, the findings of the present study suggest a more general problem in sequencing, which emerges in other functions besides speech functions. Above it is discussed that frontal lobe pathology can result in problems in planning and programming. Since frontal lobe pathology in DAS is suggested in neurological studies, the suggestion of a more general problem in sequencing (including planning and programming of speech) may indicate a possible involvement of the frontal lobe in DAS.

General conclusion

The results of the present study indicate a deviant development in sequential functioning (both sequential memory and motor functioning) that is, furthermore, significantly correlated with severity of speech involvement. Additionally, a delay in the development of other cognitive functions was found in children with DAS. This suggests that DAS is a deficit in sequencing speech movements, with an underlying general deficit in sequencing in most children with DAS.

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APPENDIX A

Descriptive data of individual children, including Language Comprehension (Reynell Test), Language production (Schlichting Test), Auditory memory, audiometrics, Maximum repetition rate (MRR), and consonant substitution in repetition of meaningful and nonsense words.

Children with DAS		Reynell language comprehension		Schlichting Language production				MRR		Meaningful utterances				Nonsense utterances					
Childnr	Age	CQ	diff age	SQ	diff age	WQ	diff age	Mem.Q	Audio	Mono syll.	tri-syll.	subcons	place	manner	voicing	subcons	place	manner	voicing
1	5-00	102	0;02	87	-0;11	86	-0;08	96	moderate	3,53	1,93	26,15%	76,47%	58,82%	23,53%	46,97%	67,74%	48,39%	38,71%
2	5-01	92	-0;05	85	-1;01	88	-0;08	89	good	4,12	3,38	23,88%	56,25%	37,50%	31,25%	63,64%	78,57%	64,29%	47,62%
4	5-01	86	-0;10	82	-1;04	88	-0;08	96	Moderate	3,70	-	21,74%	93,33%	53,33%	33,33%	50,00%	84,85%	60,61%	51,52%
8	5-03	105	0;05	82	-1;05	80	-1;01	82	Good	0,00	0,00	8,70%	66,67%	66,67%	33,33%	33,33%	50,00%	50,00%	35,00%
9	5-02	80	-1;05	73	-2;02	69	-1;06	75	Good	2,70	-	14,10%	45,45%	54,55%	36,36%				
10	5-04	99	-0;03	73	-2;02	93	-0;05	96	Good	2,48	-	10,77%	14,29%	28,57%	100,00%	18,18%	41,67%	25,00%	83,33%
11	5-05	94	-0;04	71	-2;05	92	-0;06	89	Moderate	4,61	-	39,13%	70,37%	44,44%	51,85%	68,18%	75,56%	60,00%	55,56%
12	5-05	91	-0;08	76	-1;10	87	-0;09	104	Moderate	3,02	-	20,59%	78,57%	42,86%	50,00%	43,94%	75,86%	41,38%	34,48%
13	5-06	84	-1;04	73	-2;02	72	-1;06	82	Good	3,64	2,21	21,88%	71,43%	57,14%	21,43%	38,46%	68,00%	36,00%	56,00%
14	5-07	83	-1;04	75	-1;11	85	-1;00	96	Good	4,63	3,68	14,93%	60,00%	30,00%	10,00%	15,15%	30,00%	80,00%	30,00%
20	5-11	92	-0;06	69	-2;04	89	-0;10	75	Good	3,31	-	29,85%	60,00%	25,00%	50,00%	48,48%	71,88%	56,25%	37,50%
21	5-11	89	-0;10	73	-2;02	94	-0;09	96	Good	3,20	-	31,00%	85,00%						
22	6-01	96	-0;03	67	-2;09	79	-1;05	75	Good	3,31	-	29,41%	65,00%	55,00%	35,00%	66,10%	64,10%	53,85%	30,77%
25	6-05	93	-0;08	66	-3;04	81	-1;07	93	Good	4,31	-	44,78%	76,67%	36,67%	43,33%	78,79%	73,08%	51,92%	38,46%
26	6-06	84	-1;02	66	-3;05	79	-1;09	96	Good	3,76	-	35,38%	78,26%	43,48%	30,43%	46,97%	64,52%	38,71%	64,52%
28	6-10	82	-1;08	66	-3;08	89	-1;07	89	Good	4,23	3,61	40,32%	60,00%	48,00%	48,00%	39,39%	96,15%	38,46%	38,46%
29	6-10	93	1;02	74	-2;08	100	-0;09	89	Good	3,98	-	13,85%	55,56%	66,67%	11,11%	15,15%	60,00%	60,00%	50,00%

Appendix A (continued)

NS Children		Reynell Language comprehension		Schlichting Language production				MRR		Meaningful utterances					Nonsense utterances				
Childnr	Age	CQ	diff age	SQ	diff age	W Q	diff age	Mem.Q	Audio	Mono syll.	tri-syll.	subcons	place	manner	voicing	subcons	place	manner	voicing
31	5-05									4,06	3,65	2,90%	0,00%	0,00%	100,00%	7,58%	0,00%	0,00%	100,00%
33	5-07									4,23	5,26	0,00%	0,00%	0,00%	0,00%	7,58%	0,00%	20,00%	80,00%
36	5-00									4,85	3,68	0,00%	0,00%	0,00%	0,00%	16,92%	63,64%	63,64%	63,64%
38	5-01									5,12	2,69	5,97%	0,00%	25,00%	75,00%	34,85%	56,52%	47,83%	39,13%
39	6-06									4,85	5,21	3,28%	0,00%	50,00%	50,00%	7,58%	20,00%	40,00%	100,00%
40	4-11									4,36	1,58	5,80%	25,00%	25,00%	100,00%	6,06%	25,00%	0,00%	75,00%
42	5-03									4,74	4,05	5,19%	25,00%	50,00%	25,00%				
43	6-01									4,51	2,89	1,56%	0,00%	100,00%	0,00%	3,03%	50,00%	0,00%	50,00%
44	5-03									5,64	2,35	3,17%	0,00%	0,00%	100,00%	10,61%	14,29%	14,29%	100,00%
45	5-03									4,22	-	11,54%	66,67%	44,44%	33,33%				
47	5-09									4,35	4,33	0,00%	0,00%	0,00%	0,00%				
49	5-11									4,97	3,06	1,47%	0,00%	100,00%	0,00%	12,50%	0,00%	16,67%	83,33%
51	5-07									4,71	-	4,48%	0,00%	0,00%	100,00%	16,67%	36,36%	27,27%	45,45%
53	5-08									4,49	5,03	8,47%	0,00%	60,00%	40,00%	7,58%	20,00%	20,00%	80,00%
54	4-09									4,29	2,83	7,46%	0,00%	0,00%	100,00%	12,12%	37,50%	0,00%	75,00%
58	5-06									4,44	5,08	4,69%	33,33%	33,33%	33,33%	9,09%	33,33%	0,00%	66,67%
59	6-05									4,56	4,05	0,00%	0,00%	0,00%	0,00%	10,61%	57,14%	71,43%	71,43%

Note. diff. age=age difference of the scores relative to norm-age scores; CQ=comprehension quotient (>80 is normal); SQ=sentence production quotient; WQ=word production quotient; Mem.Q=Auditory Memory Quotient; MRR=Maximum Repetition Task; monosyll=monosyllabic repetition (/pa/, /ta/, and /ka/), trisyll=trisyllabic repetition (/pataka/); Subcons=percentage singleton consonant substitution syllable initial; place/manner/voicing=percentage substitution of place/manner/voicing of subcon. Missing data are indicated with blank cells.

APPENDIX B

Detailed description of subtests.

1. Motor functioning

a. Motor execution *Finger tapping*: pressing a telegraph key with the index finger of one hand as many times as possible in ten seconds. This is repeated five times, and the average number of tapping is calculated. This subtest is derived from a Dutch neuropsychological testbattery (P.I.N.O.K: Neuropsychologisch onderzoek), however, age norms are only available from the age of 6 onwards.

b. Motor planning Psycho-motor planning *Labyrinths* (RAKIT): going through a labyrinth with a pencil as fast as possible. This is a visual motor test, in which perceptual integration, planning, and speed are of importance. Cumulative scores are determined, in which the time it took to pass a labyrinth is transposed into a score; the longer it took the lower the score. (age norm scores: M=15, sd = 5).

Sequential motor planning *Hand movements* (K-ABC): performing a series of handmovements (increasing in complexity of three hand positions) in the same sequence as the examiner performed them. Scores consists of responses in which both sequence and number of hand movements were correct. (age norm scores: M=10, sd = 3).

Auditory motor planning *Auditory rhythm*: a rhythm tapped with one hand is presented auditory and has to be repeated. After that, a rhythm is tapped with alternating movements of both hands, which is presented both auditory and visually, and has to be repeated. Scores consist of correct responses of the preference hand, the non-preference hand, and both hands.

2. Memory

a. Sequential memory *Number recall* (K-ABC): digits that are presented auditory have to be remembered and enumerating in the right sequence. The number of correct sequences are counted. (age norm scores: M=10, sd=3).

Word order (K-ABC): words that are presented auditory have to be remembered and the corresponding pictures have to be pointed out in the right sequence on a larger illustration. The correct responses (in the right sequence) are counted. (age norm scores: M=10, sd=3).

b. Simultaneous memory *Spatial memory* (K-ABC): the location of the pictures have to be remembered and pointed out. Correct responses are counted. (age norm scores: M=10, sd=3), norm scores only available from the age of 5 onwards.

3. Sensory

a. Sensory *Oral sensory*: a three-dimensional object has to be identified in the mouth and the corresponding picture in a row of five pictures has to be pointed out. Correct responses are counted.

Finger localization: the child has to indicate which finger (or fingers) has been touched, with and without looking during touching. Scores consist of the correct responses of one-finger touching with visual information, the correct responses of one-finger touching without visual information, and the correct responses of two fingers touched sequentially again without visual information. This subtest is derived from the description in Benton, Hamsher, Varney, and Spreen (1983). There are no aged norm scores available, only average scores are reported (p.92).

4. Attention

Deutsche Schulkonzentrationstest: figures of pears have to be crossed out (per row) on a paper with rows of apples and pears. Scores consists of the average time needed for the execution per row and number of omissions.

CHAPTER SEVEN

General Discussion and Conclusion

INTRODUCTION

The diagnostic features that may discriminate children with developmental apraxia of speech (DAS) from other speech disorders are argued about in the research to DAS, due to an overlap in characteristics (McCabe, Rosenthal, & McLeod, 1998). Besides aiming at finding a specific diagnostic marker for DAS, research concerning DAS focuses at trying to detect the underlying deficit of DAS (see General Introduction). Knowledge of the underlying deficit leading to DAS will enlarge our understanding about the disorder and more specifically about differential diagnostics. Starting point of the research presented in the present thesis was the question where in the speech production process the deficit of DAS might be located. This study was based on results of previous studies that were carried out in the Department of Pediatric Neurology (University Medical Center Nijmegen) in order to objectively describe the speech characteristics of DAS (Thoonen, Maassen, Gabreëls, Schreuder, & De Swart, 1997; Thoonen, 1998). A key result of these previous studies was the development of a series of assessment procedures to diagnose children with DAS (from other speech disorders, such as specific language impairment and dysarthria). Using these procedures, the severity of involvement was quantitatively indexed showing the data that DAS is a matter of degree; many children with speech disorders (of divergent origin) to some extent show dyspraxic characteristics (Thoonen, 1998).

The present thesis builds on this knowledge making the following step: in addition to assessing the degree of involvement, the location of the underlying deficit in DAS within the speech production process was investigated. More specifically, knowing what the clinical symptoms of DAS are, we investigated underlying information and speech processing deficits. Hereto a specific study design was formulated that consists of the following parts. Firstly, as starting point we used a model of speech production (see General Introduction; this model was based on Levelt's model, 1999, and extended for the more motoric part based on the model of Van der Merwe, 1997). Subsequently, based on this model of speech production and the characteristics of DAS several hypotheses on the underlying deficit were formulated in which we incorporated children with DAS having problems in phonetic planning and/or motor programming of speech production. The involvement of each successive level of speech production in DAS was systematically investigated in different experiments.

An important aspect of the study's design was the selection of children with DAS. Stringent selection criteria were used to select a group of children with DAS that could be considered clear cases, avoiding additional problems such as language comprehension problems, dysarthria, hearing problems, and below average intelligence. In the experiments, utterances of children with DAS were compared to those of normally speaking children. (Since both normally speaking children as well as children with DAS of the ages studied in the present study (4;11 to 6;10 years), are in a process of maturation and development of their speech motor system, the experiments were also conducted with adult women to obtain reference data on the mature speech motor system.) It might appear to be trivial that the results of children with DAS were expected to be different from those of normally speaking children, but since the present study concerned a more fundamental question, comparison with a normally developing speech motor system was essential. The present study aimed at establishing possible problems in successive levels of speech

production in children with DAS, which cannot be determined during clinical diagnosis. Therefore, we investigated possible deviancies in phonemically correct utterances of both children with DAS and normally speaking children, rather than collecting and counting the number of phonemic substitutions.

A summary of the results of the previous chapters reveals that the answer to the fundamental question whether children with DAS show a deficit at the level of phonetic planning and/or speech motor programming is clearly positive: both planning and programming are deviant. As part of the planning deficit, also deficits in 'higher' mechanisms, concerning durational control processes and use of prosody, seem to be at play in DAS. As part of the programming deficit, we also found evidence for problems in the process of automating speech production. A more generalized problem in sequencing and timing is suggested to be underlying these two deviant processes. From a neuropsychological point of view, these speech motor planning, programming, and automation deficits appeared to be embedded in more general information processing deficits, such as deficits in sequencing abilities and sequential memory. The latter results, moreover, suggested a more generalized problem in sequencing and timing abilities.

Before elaborating on this suggestion of a more generalized problem, we will first summarize and discuss the main conclusions of the previous chapters.

RESULTS OF THE EXPERIMENTAL STUDIES

The results of Chapter 2 showed that the normally speaking children exhibited more inter-syllabic coarticulation than the adult speakers, which indicates that during speech development the articulation becomes more and more segmented. This suggests that coarticulation – or the lack of coarticulation – is an important parameter to assess developmental speech disorders and to evaluate the involvement of planning and/or programming of speech movements in context. Children with DAS showed both quantitative and qualitative differences in coarticulation, that is stronger coarticulation and idiosyncratic patterns, as compared to normally speaking children, suggesting delayed as well as deviant speech development. In particular, the results could be explained as arising from a deficient automation of speech production. In addition, children with DAS showed more variability in repeated utterances than the normally speaking children, which is an indication of less mature automation (i.e., 'younger' speech).

Evidence for a deficit in phonetic planning of syllables in children with DAS was found in Chapter 3. Whereas systematic durational adjustments to the metrical structure of the syllable were found in the segments of the stressed syllable in normally speaking children, in children with DAS such systematic durational patterns were missing. This suggests that during speech production in children with DAS each single segment is processed on its own, without accounting and adjusting for surrounding segments within the syllable. Furthermore, the overall timing patterns suggested that children with DAS do not process prosodic properties similarly as normally speaking children do. Again, high within-subject variability was found in children with DAS, indicating a less automated speech production process, especially in motor programming.

Evidence for a possible deficient use of the syllabary in children with DAS could not be shown in experiments that manipulated syllable frequency; nor was an effect of syllable frequency

observed in normally speaking children. Thus, there was no evidence for the existence of a syllabary, as suggested in the initial model.

Problems in (automating) motor programming in DAS, as suggested with the variability in repeated utterances in Chapter 2 and 3, emerged in a bite-block speech condition in children with DAS (Chapter 4). Although neither normally speaking children nor children with DAS were able to completely compensate for the bite-block, the bite-block did not affect the extent of anticipatory coarticulation in normally speaking children. In this respect, the normally speaking children showed similar results as the adult women. In contrast, in the speech of children with DAS large effects of the bite-block were found on vowel quality, which, contrary to expectations, had improved. This result was interpreted as a clear demonstration of deficient motor programming in DAS, in a less automated and controlled processing mode.

The durational results of Chapter 5 revealed a lack of significant contextual interdependency in children with DAS as compared to normally speaking children in a normal speech condition, which suggested a possible lack of durational control. The bite-block speech condition corroborated this suggestion of deviant durational control, which furthermore supports the suggestion made in Chapter 3 that children with DAS consider every segment on its own without adjusting for the phonetic context. Thus, when combining the results of Chapter 5 with those of Chapter 3 we may conclude that a lack of durational control affects phonetic planning (i.e., a lack of syllabic durational structure) as well as motor programming (i.e., a lack of adjustment to phonetic context in both a normal speech condition and a bite-block speech condition).

Finally, in Chapter 6, results of the cognitive tests administered to children with DAS indicated a deviant development in sequential abilities (both sequential memory and motor functioning), which were significantly correlated with severity of speech involvement. The children with DAS showed deviant sequential abilities in a background of overall lower scores. They, however, showed a similar improvement during development as compared to normally speaking children, which indicated a delay in development of about 15 months. When relating these results, indicating a general deviance in sequential abilities, to the speech production model we have further evidence for problems in phonetic planning and motor programming in children with DAS.

In the present study we investigated a small group of children, who were selected using stringent criteria. Despite this small group we still found significant effects. Because of the stringent selection criteria we were able to investigate a group of children with the most clear diagnosis of DAS as one can get. These stringent criteria plus the experimental design, in which each successive level of speech production was investigated separately, allowed us to draw firm conclusions about specific deviancies in underlying processes. In the literature, the views about the underlying deficit of DAS are divided, partly due to heterogeneous groups that are investigated and the lack of experimental studies that explicitly investigated each level of speech production separately. Thus, the results of our studies yield a valuable contribution in the debate about the underlying deficit in DAS. Instead of speculating about underlying deficits based on clinical symptoms, we are now able to substantiate underlying deficits due to stringent selection criteria and systematic investigation. Nevertheless, did our data provide an unambiguous answer as to the question what might be the underlying deficit in children with DAS?

IS THERE ONE UNDERLYING DEFICIT?

As was stated in the General Introduction, children with DAS generally present symptoms with various deficits: besides speech problems (low intelligibility due to inconsistency in consonant substitutions) also soft neurological signs and clumsiness are mentioned (an overview of features of DAS is given by McCabe et al., 1998). Various researchers discussed the possibility that the speech output of children with DAS reflects a compensation strategy for a more general disability. This means that the possibility of a more general disability should be considered. And subsequently, it is important to determine for each symptom whether it directly reflects the underlying disorder or is more likely to be the result of a compensation strategy. On the one hand, if a symptom is the result of normal processing then this can be understood as a compensatory symptom. That is, a compensatory symptom is the result of adapted functioning of a normal mechanism under abnormal circumstances or in the presence of a proved disorder at another level. For instance, distorted grammar in telegraphic speech resulting from a short-term memory deficit is a compensatory symptom since the grammatical knowledge can be considered to be unimpaired (Kolk, 1995). On the other hand, if a symptom could only be explained as the result of deviant mechanism then it must be explained as a symptom of the disorder. This issue of compensatory symptom versus symptom of the impairment is in particular relevant with respect to suprasegmental aspects of speech. It is, for instance, hard to distinguish whether slow speaking rate is a symptom of the disorder or a symptom of compensation (also see Chapter 5). Davis, Jakielski, and Marquardt (1998) reported suprasegmental abnormalities in DAS, however, they stated that it is not clear whether these abnormalities are a part of the disorder or a compensation for the impaired ability to produce syllable sequences. Shriberg, Aram, and Kwiatkowski (1997a) showed that children with DAS experience difficulties in using stress. In contrast to Davis et al. (1998), Shriberg et al. (1997b) were more explicit and suggested that this difficulty in using stress reflects a problem at a higher level of speech production instead of a compensatory effect.

On the basis of these clinical studies, however, it is impossible to decide whether the suprasegmental abnormalities, such as slow speaking rate and inappropriate use of stress, are resulting from compensatory processing for a deficit at a different level, or from a primary suprasegmental planning deficit. The above-mentioned studies suggested diverse deficits in DAS. Yet another option was proposed by Marion, Sussman, and Marquardt (1993), who discussed the possibility that the deficiencies in speech motor output reflect an ill-formed phonological representation system. Furthermore, Dodd and McCormack (1995) mentioned that it is difficult to substantiate whether the poor performance of children with DAS on tests of receptive and expressive vocabulary and motor planning for verbal and non-verbal tasks reflect a number of different and distinct underlying deficits, or feedback and feedforward effects from a single deficit. Thus, various studies suggest planning problems in higher linguistic processes in children with DAS (see also Hodge, 1994; McCabe et al., 1998; Velleman & Strand, 1994), which are in accordance with the clinical observations and which indicate comorbidity.

Besides this issue of a deficit at a different level of linguistic processing there might be a more general impairment. Velleman and Strand (1994; p.119-120), for instance, suggested a possible common underlying information processing factor that children with DAS “could be

seen as impaired in their ability to generate and utilize frames, which would otherwise provide the mechanisms for analyzing, organizing, and utilizing information from their motor, sensory, and linguistic systems for the production of spoken language". This was interpreted as follows: children with DAS "might 'have' appropriate phonological (or syntactic) elements but are unable to organize them into an appropriate cognitive hierarchy" (p.120). Combining the results of the previous chapters of the present thesis, especially with respect to the deficits in sequencing abilities that were found in the last experiment of the present thesis, that were, furthermore, correlated with the speech characteristics, we also suggest a general problem in DAS. That is, the results of the experiments described in the current thesis could well be explained in the light of a more generalized deficit in sequencing and timing, that emerges in speech motor control, phonetic planning, and motor programming, and also in sequential memory and sequential movements. Possibly there is a common (neurological) substrate that might account for deficits in these areas.

Sequencing and timing are two aspects of a process that are hard to separate. Metaphorically these aspects could be compared with an orchestra performing a composition (in which each member of the orchestra uses the same musical notation as metaphor for a correct phonological plan). Both the sequence and the timing of the instruments that are played are important. If the instruments are not played in the correct sequence the music piece will sound awkward. But also the timing by which the successive instrument are played, requires conscientiousness. If not, some instruments may follow the others too quick, whereas others may be delayed. This will then result in inconsistent tempos and the composition will sound distorted. The performance of the composition by the orchestra stands or falls with the competence of the conductor to point to the successive instrument players in the right sequence and with the right timing. As in the orchestra, also in speech production sequencing and timing are very important. There are different 'instruments' involved in the production, that is, the speech organs jaw, lips, tongue, vocal cords, and respiration organs. And the movements of the organs (the contraction of the muscles in question) must be planned and programmed in the right sequence and the right timing (interesting is the fact that neurological conduction, from neurological command to muscle movement, is different for the different speech organs; the massive jaw is notoriously slow). The speech characteristics of DAS as well as results of the experiments presented in this thesis suggest a generalized problem in sequencing and timing in DAS. Below, we sum up evidence that corroborates the suggestion of such a generalized problem.

First, the high number of phonemic errors that are commonly found in DAS might be a result of a problem in sequencing, in that the incorrect sequence of phonemes was selected, or that the correct sequence could not be retained (in our metaphor, this indicates that the musical score is also distorted). Moreover, qualitatively the pattern of phonemic errors, in which place of articulation appears more frequently than other errors, is different from normal error patterns. Errors in place of articulation could well be explained by problems in sequencing the right articulatory movements (Kent & Rosenbek, 1983). The same applies for the problems in diadochokinesis, in which the children with DAS have trouble sequencing the correct syllables (/pataka/) and when successful in producing the correct diadochokinetic sequence they show longer syllable durations than normally speaking children. Second, the groping behavior (i.e.,

searching articulation behavior) that is reported in most studies of DAS is another indication of a sequencing and timing deficit, concerning the control of articulatory movements. Third, a sequencing and timing problem emerges in the incorrect use of stress and in scanning speech, in that all syllables in a word and all words in a phrase are stressed. These are characteristics of DAS that are frequently reported and could be explained as an inability in timing and sequencing the correct stress pattern. Fourth, the results of the compensatory speech for a bite-block (which indicated motor programming problems) might be explained as resulting from a sequencing and timing problem, in that the correct articulatory movements have to be quickly programmed and coordinated in the right sequence. Fifth, also the lack of contextual interdependency as found in the segment durations within syllables reflects sequencing and timing problems. The segment durations were not adapted to the durations of surrounding segments or to metrical changes within the syllable. The common underlying principle seems to be that children with DAS treat each part of their performance, in whatever domain, as a separate entity. They do not merge (fluently) and they do not automate. Apparently, the segments are treated separately without accounting for the contextual segments. Finally, the weaker sequential memory and sequential motor abilities as were reported in Chapter 6 comprised the major evidence for a more generalized problem in sequencing.

Thus, in the present study we found suggestions for one underlying deficit in DAS that affects sequencing and timing. However, a timing and sequencing deficit is also suggested in other speech-language disorders, which makes it aspecific for DAS. Various authors have suggested that the underlying deficit which leads to speech and language disturbances is one of processing of timing in both the perception and production (Hammond, 1982; Katz, Kripke, & Tallal, 1991; Wolff, 1993). Timing is thought to be a necessary component in tasks involving coordinated actions. Tallal et al. (1998) investigated the perception of very short intervals and rapid acoustic changes and explained perception disorders as an inability to process very fast stimuli in perception. Other studies reported impairments in perception and production of rhythm tasks (with longer intervals), which were interpreted as impairment in patterns of timing (e.g., see Alcock, Passingham, Watkins, & Vargha-Khadem, 2000). Furthermore, Miller (1989) suggested a relative timing deficit in dyspraxia. Also, Ziegler and Von Cramon (1986) found deficient timing in acquired apraxia of speech. In a recent study, Alcock and colleagues (2000) proposed a sequencing deficit combined with a fine-grained timing deficit in adults who suffer from an inherited developmental speech-and-language disorder. Franz, Zelaznik, and Smith (1992) emphasized that there are some general mechanisms that are shared across different effector systems, that is in the movements of the limbs and the oral motor system.

If such a general problem in sequencing and timing is underlying different speech and language disorders, what, then, is the reason that some children end up with a phonological disorder, or with stuttering, and others with developmental apraxia of speech? The developmental perspective of speech disorders may explain why a general problem in sequencing and timing that is underlying different speech and language disorders expresses itself in different ways.

Developmental perspective

In the literature on clinical diagnosis the discussion concentrates on the issue whether DAS must be viewed as a syndrome characterized by a symptom cluster, or as a specific, unitary disorder with a constant physical sign referring to a single underlying deficit (Shriberg, Aram, & Kwiatkowski, 1997b). Both types of diagnostic reasoning refer to an underlying deficit. In the syndrome view the common factor is of etiological nature, in which the research question concentrates on what children with the clinical diagnosis DAS have in common as regards the origin of their speech deficit. In the unitary disorder view, the research question could be turned around: Under the assumption that the underlying deficit in DAS is a disorder in speech motor planning (McNeil, 2001), the question becomes one of defining the clinical population belonging to this underlying deficit. Whatever viewpoint one adopts, the developmental perspective must be taken into account (Hodge, 1994; Maassen, 2002).

Infant speech develops from senso-motoric learning and random babbling to more abstract phonological acquisition. The first year of speech development is dominated by motor learning, which remains important, but is gradually replaced by a more phonological orientation (Smith, 1984). Note that infant speech development consists of vocal play and babbling (before 'real' speech). Many studies have demonstrated the prognostic value of babbling in delayed or pathological speech development (e.g., Oller, Eilers, Neal, & Schwartz, 1999). During development the more or less randomly generated babbling movements of the articulatory organs form the opportunity for the child to discover the auditory effects of the articulatory system (articulatory-to-auditory mapping). The speech model of Guenther and colleagues (1998) emphasizes the importance of perceptuo-motor aspects of the developmental and clinical observations. The stages of the speech production model of children may change over time. Guenther et al. (1998) suggested that during development the speech production processes change, in that the articulatory-to-auditory mappings later are extended with imitation of speech sounds of others (auditory-to-articulatory mapping or inverse modeling), to which finally phonemic categories of the native language are added (auditory-to-phonemic mapping and phonemic-to-auditory mapping).

Thus, during speech development children develop gestures and learn to fine-tune syllabic gestural scores. If we would adapt our initial model (see Introduction) to these developmental aspects, this will result in the model shown in Figure 1, in which at first, during babbling (before 'real' speech), phonological encoding and phonetic planning are no part of the production process.

Bishop (1997) formulated a similar developmental mechanism: "there is ample evidence that the nature of underlying representations may evolve in the course of development, and there may be far more interaction between levels of processing in children than adults. (...) one needs to be aware that the profile of impairment can change with age and that an impairment affecting one stage of processing may have bottom-up or top-down influences on the development of other stages." (p. 919). This underlines the complexity of speech production processes in children with developmental speech disorders, since deficits at one level of speech production might induce a problem at another level later in development. For example, a disorder at auditory processing

might induce problems in motor programming in that a possible auditory mapping cannot be realized.

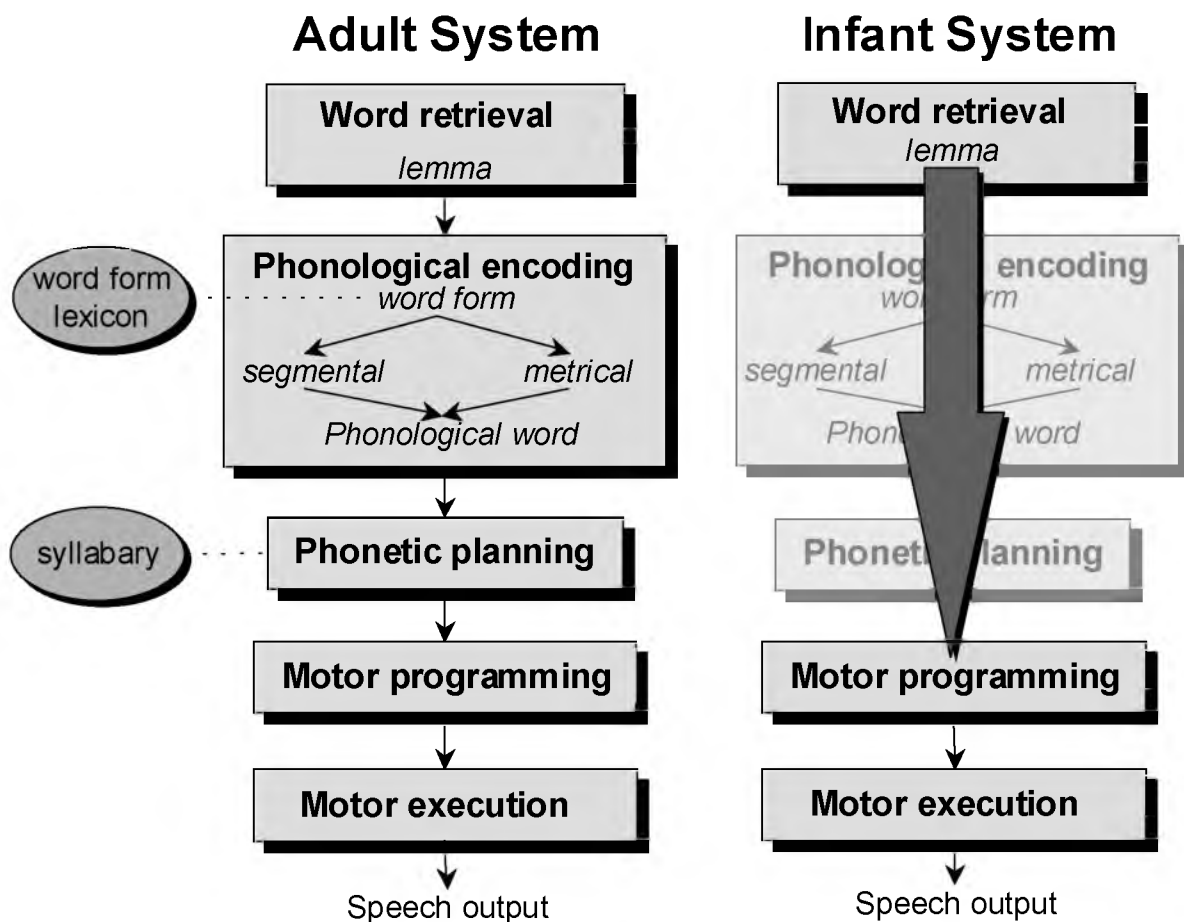


Figure 1. Adult and infant model of speech production. Infant speech development consists of vocal play and babbling (before 'real' speech). The infant model is an adapted version of the adult model accounting for changes during development.

Many authors have noted that the symptom cluster of DAS changes over time (for example see Shriberg et al., 1997b). Among the first signs of a dyspraxic development, often assessed in retrospect, reduced babbling in combination with a delayed or deviant oral motor development is found. Although this is not an exclusive characteristic, that is, other developmental speech disorders also show reduced babbling, there is strong evidence that delayed or deviant motor development and perceptual motor learning play a role in many children with DAS. Secondly, besides reduced babbling, the high incidence of concomitant dysarthria and oral motor dyspraxia (as already noted in early studies by Rosenbek & Wertz, 1973; Yoss & Darley, 1974) further suggests a generalized problem in motor control of the speech mechanisms. The third clinical evidence comprises the high comorbidity rate of DAS, especially with regard to fine and gross motor impairment (Ferry, Hall, & Hicks, 1975; McCabe et al., 1998). Thus, the symptom complex of children with DAS consists of a core set clearly referring to the underlying

impairment of timing and sequencing (namely groping behavior, incorrect use of stress, consonant substitutions in place of articulation), as well as variable symptoms.

Oral motor development and automation might explain why a similar underlying impairment in sequencing and timing results in DAS, or stuttering, or phonological problems. All children go through a stage of having to learn/acquire sequencing and timing. This acquisition process may be accompanied with weaknesses in or stress from other systems. Different developmental trajectories are possible, depending on severity, onset, and 'location' of other deficits (emphasis on motor behavior, perception, or phonetic planning). For instance, a late onset of a moderate timing deficit might lead to stuttering, in that basic articulatory patterns are acquired, but the production system is stressed because of the enormous increase of complexity imposed by the grammatical development. DAS may, then, mainly be the result of motoric timing deficits that manifest themselves at a young age (with low complexity of utterances), and subsequently frustrate phonological development (symptoms change during development).

IMPLICATIONS FOR FUTURE RESEARCH

Although scientific research reveals valuable answers to research questions, it also produces new (additional) questions to be investigated. Therefore, a few implications for future research are mentioned below.

Models of speech production provide us with a useful framework to investigate development of speech production and pathological speech production. However, we have to realize that these models are composed on the basis of normal speech production in adult speakers. The use of these models in interpreting pathological and developmental speech production has to be taken with some precaution. As mentioned above, the stages of the speech production model of children may change over time (also see Figure 1). Therefore, future research should also focus on speech production (including babbling) in younger children (than the ones in the present study), which will further broaden our knowledge on development of speech production and on problems that might occur at various stages of development and levels of the speech production process. One must be aware of interactions in development. The result of the longitudinal study, in the present thesis, suggests that following children during development reveals valuable information about delay and deviance. Information at an even younger age, during the babbling phase (before 'real' speech production), might further recognize deviancies in developmental patterns.

Additionally, perception plays an important role in motor learning and automation (Guenther, Hampson, & Johnson, 1998). And therefore, research on how children with DAS and normally speaking children learn new phonemes or clusters of phonemes in production as well as perception will further enlarge our knowledge on acquisition of speech and the problems that might be induced during development.

In the present study, we investigated children with DAS and compared their results with those of normally speaking children. Since we did not investigate children with other speech disorders, or children with specific language disorders, we are not able to draw conclusions as to the specificity of the findings for DAS. Nevertheless, the results of the present study might also

be relevant for other speech disorders. Especially the notion of a generalized mechanism of timing and sequencing underlying DAS and the apparent problems in timing and sequencing in other speech disorders require further investigation in other speech and language disorders. Moreover, extensive research on various clinical research populations will enable us to further specify the findings for DAS. Furthermore, it would be interesting to investigate the timing and sequencing problems in speech disorders (of various origin) in a more direct way by studying coordination of speech movements (of the articulators) rather than the acoustic output. Motor coordination (in which timing is important) can be investigated using Electromagnetic Articulography by which the articulatory coordination between the articulators (the lips, tongue tip, tongue body, and jaw) can be determined.

Results of the present study showed that children with DAS have problems in timing and sequencing. Furthermore, the syllable plays a central role in speech production (including timing and sequencing), also in children with DAS. On the basis of these results suggestions for therapy can be made. One training program for children with DAS is the “Nuffield Dyspraxia Program”, which has been translated and modified for Dutch (Erlings-van Deurse, Freriks, Goudt-Bakker, Van der Meulen, & de Vries, 1993). This therapy program is an example of how to implement some of the basic principles: from simple to complex articulations (based on simple, basic overlearned patterns) and flexibility in combinations. The use of this program is widespread in the Netherlands. However, the literature also mentions other suggestions for therapy in DAS (and adult apraxia). Therapy methods for the treatment of (adult) apraxia of speech are mentioned that are directed towards facilitating the positioning of the speech musculature and/or the coordinated phasing of speech subsystems at the segmental and syllable level (Square-Storer, 1989), such as shaping speech sounds from nonspeech sounds, gradually shaping speech segments from other speech segments, or extending from key words to other words comprising the same phonemes. Furthermore, Square-Storer (1989) described a few methods of treatment facilitating the temporal schemata of speech and the sequencing of segments in longer utterances. These methods comprised melodic intonation therapy (MIT), based on melodic line, tempo, intersystemic reorganization, in which a more or less intact motor system (e.g., finger tapping) is paired with another less intact one (speech motor), and the PROMPT system (Prompts for Restructuring Oral Muscular Phonetic Targets), in which intersystemic reorganization is combined in spatial and temporal aspects of speech production (Chumpelik, 1984). Therapy programs like the above, which are based on clinical experience rather than fundamental research, form an interesting starting point for further therapy evaluation.

With the knowledge obtained in the present study improvements can be suggested and implemented in therapy programs that should be further investigated and therapy should be evaluated. First, as stated above, early oral motor and speech production development seems to be important in the development of a speech disorder, especially DAS. This indicates that it is very important to start with therapy at a young age (which was also emphasized by Noterdaeme, Mildemberger, Minov, & Amorosa, 2002). Enhancing oral motor abilities in babbling or speech like tasks early in development will have a positive influence on later stages of development. Second, the results of the present study suggest that the syllable should be the basis for therapy, rather than the isolated segment, in order to improve coordinated motor patterns of the size of a

syllable. Third, contrasts between syllables might be used to learn different phonemes. This forms an imitation of normal development. By doing so, sequencing abilities are trained simultaneously with learning contrasts between phonemes. Fourth, training with alternating intonation patterns in words (and later in phrases) will improve timing abilities. Finally, the motor patterns should be trained over and over again.

Besides speech therapy it might be interesting to evaluate whether improving sequencing and timing in other systems (e.g., concerning general motor functioning or sequential memory functioning) will have a positive effect on sequencing and timing in speech production.

Evidence for a neurological problem in DAS is still not found. Brain imaging techniques are developing quite rapidly. Location of a neurological problem in DAS and event related neurological deviancies in DAS might enhance our understanding of the disorder, especially in the comparison with other disorders (acquired and developmental). Furthermore, brain imaging techniques, such as ERP and fMRI, might reveal indications of a location for sequencing and timing, and through this indications for a possible neurological problem in DAS.

The present thesis showed that using stringent selection criteria of children with DAS remains important. Besides using stringent selection criteria we have to reach international agreement on the criteria. Proposals for consensus on this issue are suggested in the past, however, worldwide consensus is still lacking.

In conclusion, the time seems to be ripe for a research program consisting of stringent diagnostic criteria (with international agreement), further study of the underlying disorder in which also early development and motor coordination should be investigated, brain imaging techniques (such as ERP and fMRI), and therapy evaluations.

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Summary

Speaking appears to be a simple task for those who do not experience problems while producing speech. However, if you have ever worked with children or adults with a language or speech disorder you will know that this ability cannot always be taken for granted. This thesis deals about children with developmental apraxia of speech (DAS) for whom producing intelligible speech is very hard. Their utterances contain a large number of sound errors and distortions. Although the disorder is classified as a neurogenic speech disorder, to date no clear neurological deficit has been demonstrated in children with DAS. Likewise, cognitive studies searching for the problem in the speech production process were as little successful as to find an unanimous answer concerning the underlying deficit of DAS within the speech production process. The ultimate goal of the present thesis was to find an answer to this question of underlying deficit in the speech production process. For this we used a model of speech production and tested the involvement of successive levels of the speech production process in the underlying deficit of DAS.

The Introduction (chapter 1) briefly describes the speech characteristics of DAS, followed by a clarification of the stages in the speech production model that we used. Subsequently, we formulated hypotheses about the stages of speech production that might be impaired in children with DAS, namely phonetic planning (including the idea of using a syllable repository), motor programming and motor execution. These hypotheses were tested in experiments described in the chapters 2 to 6. By means of acoustic analyses, that is second formant (F2) values and segment durations, the influence of the phonetic environment on the production of segments (i.e., coarticulation) was investigated in utterances produced by normally speaking children and children with DAS.

The hypothesis of the first experiment, described in chapter 2 ('Coarticulation patterns'), was based on an aspect of speech development that in normal development younger children show more coarticulation in their utterances than older children. This implies that young children start with more global articulation than older children in which differentiation in phonetic segments is not yet realized. During speech development the articulation gets more and more segmented. This suggests that the phonetic segment is the endpoint rather than the starting point of speech development, and, furthermore, that the extent of coarticulation – or the lack of coarticulation – is an important parameter to assess developmental speech disorders. The first experiment studied coarticulation patterns of normally speaking children and children with DAS. This study consisted of a rather simple speech task, in which children (both normally speaking children and children with DAS) and adult women were asked to repeat nonsense CV-syllables such as 'ba', 'di', and 'su' in a carrier phrase ('hé de CV weer' [he də CV wɪ:r]). The results showed that the normally speaking children exhibited more inter-syllabic coarticulation than the adult speakers, which indicates that the assumption the study was based on (viz. coarticulation decreases during development) is correct: thus, younger speech can indeed be characterized as being more coarticulated.

In comparison to the normally speaking children the children with DAS showed more variability in repeated utterances, made, on average, less distinction between the vowels, and

showed different coarticulation patterns consisting of stronger intra-syllabic coarticulation. Furthermore, idiosyncratic patterns were found in the individual children with DAS. The quantitative difference in coarticulation was interpreted as a sign of speech delay (stronger coarticulation in younger speech), but because of the idiosyncratic patterns found in the children with DAS, we concluded that speech motor development in these children is also deviant. In particular, the results could be explained as arising from a *deficient automation* of speech production. The higher variability in repeated utterances as found in the children with DAS corroborated this indication of less mature automation (i.e., ‘younger’ speech). Thus, children with DAS show both quantitative and qualitative differences in coarticulation as compared to normally speaking children, suggesting *delayed* as well as *deviant* speech development.

Chapter 3 (‘Planning of syllables’) describes the experiment that investigated the possibility of a deficit in phonetic planning of syllables in children with DAS. For this, the syllable structure was manipulated in an otherwise unchanged phonetic context (resulting in Dutch phrases like ‘zus giet’ versus ‘ze schiet’ cf. in English ‘ice cream’ versus ‘I scream’). We expected to find a strong intra-syllabic effect of syllable structure on coarticulatory cohesion and durational structure (this means that in the English example above, the [s] will show more intra-syllabic coarticulation of the [i] in the phrase ‘I scream’ than in ‘ice cream’). Intra-syllabic coarticulation results, as measured in formant values, showed that the syllable structure did not have a systematic effect in neither children with DAS nor normally speaking children. Durational patterns, on the other hand, showed a syllable structure effect in the second syllable, but only in the normally speaking children. This means that systematic durational adjustments were found in the segments of the second syllable in that the durations of the consonant as well as the vowel were shorter in case the /s/ was added to the second syllable (i.e., in ‘schiet’ versus ‘giet’). The absence of such systematic durational patterns in the speech of children with DAS was interpreted as a deficit in phonetic planning, that is a lack of durational adjustment to the *metrical structure* of the syllable. This suggests that in the speech production of children with DAS each single segment is processed on its own, without accounting and adjusting for surrounding segments within the syllable.

Furthermore, inter-syllabic effects were found in coarticulatory cohesion as well as durational structure, due to prosodic differences between the phrases. In normal speech, phonemes in unstressed syllables are more likely to be coarticulated and have shorter durations than the same phonemes in stressed syllables. This effect was found in the utterances of the normally speaking children, that is the schwa in ‘ze schiet’ (unstressed) showed more coarticulation and was shorter than the schwa in ‘zus giet’. This effect was absent in the speech of children with DAS. The lack of such a prosodic effect on inter-syllabic coarticulation and segment duration of the first vowel, furthermore, supports the hypothesized problem in phonetic planning. This suggests that children with DAS do not process *prosodic properties* similarly as normally speaking children.

Again, high within-subject variability was reported in children with DAS, indicating a less automated speech production process, especially in motor programming. Evidence for a possible deficient use of the syllabary in children with DAS could not be shown in experiments that manipulated syllable frequency; nor was an effect of syllable frequency observed in normally

speaking children. Thus, there was no evidence for the existence of a syllabary, as suggested in the initial model.

The variability in repeated utterances, as is described in chapter 2 and 3, suggested that children with DAS have problems in (automating) motor programming. In chapter 4 ('Evidence of motor programming deficits'), a possible deficit in motor programming was further evaluated, using a bite-block speech condition. Although the normally speaking children were not able to completely compensate for the bite-block (the F2 values were higher in the bite-block condition as compared to the normal speech condition) speaking with a bite-block did not affect the extent of anticipatory coarticulation. In this respect, the normally speaking children showed similar results as the adult women. The children with DAS were neither able to compensate for the bite-block, however, in these children speaking with a bite-block led to lower F2 values. Furthermore, in the speech of children with DAS large effects of the bite-block were found on vowel quality, which, surprisingly and contrary to our expectations, had improved. This result seemed to suggest that children with DAS were actually helped by the bite-block. Probably, the restriction in degrees of freedom of the articulators facilitated articulation in children with DAS in that less aspects of the articulatory movements have to be controlled. However, an increase of within-subject variability in the bite-block condition indicated that the children with DAS, even though the vowel quality improved, did experience difficulty in the articulatory process while speaking with a bite-block. These results were interpreted as a clear demonstration of *deficient motor programming* in DAS, in a less automated and controlled processing mode.

Durational control in children with DAS was investigated in chapter 5, to evaluate whether slow speaking rate might be a compensatory symptom or the direct result of the disorder itself. For this, durational patterns, that is intrinsic and relative durational characteristics due to contextual interdependency, were compared between children with DAS and normally speaking children in two speaking condition, a normal speech and bite-block condition. The normal speech condition showed strong contextual interdependency in the segments of the normally speaking children. That is, differences in intrinsic durations between plosives and fricatives (/s/ and /x/ were longer than /b/ and /d/), and between low and high vowels (/i/ was shorter than /a/ and /u/), were compensated for by the following vowel, or preceding consonant respectively. This systematic effect of intrinsic durational differences and contextual interdependency was not found in children with DAS. This result, together with high durational variability, suggested that durational control was less strict in children with DAS.

Durational control mechanisms, especially concerning compensatory strategies, were further investigated by comparing the durational patterns in a normal speech and bite-block speech condition. The results showed that in normally speaking children, despite an increase of segment durations in the bite-block condition, the durational pattern remained stable (both vowel and consonant duration increased linearly). Thus, the normally speaking children used a compensatory strategy by slowing down speech production while controlling for the intrinsic durational differences and contextual interdependency by maintaining the durational pattern. Children with DAS showed a different mechanism to adapt to the bite-block, that is the consonant duration increased whereas the vowel duration decreased. These results corroborated the suggestion of *deviant durational control* as underlying the speech production of children with

DAS. Furthermore, these results support the suggestion made in chapter 3 that children with DAS control each segment on its own with adjusting for the phonetic context. The lack of a 'higher' durational control mechanism can well explain these results. The speech production model does not specifically mention a durational control mechanism, although, when combining the results of chapter 5 with those of chapter 3, we may conclude that durational control is relevant in phonetic planning (segment duration depends on syllable structure) as well as motor programming (longer segment durations due to bite-block but unchanged durational patterns). Thus, a lack of durational control in DAS affects both phonetic planning and motor programming.

Chapter 6 ('Cognitive functions') described the results of several non-speech tasks that were conducted in order to investigate whether the children with DAS, although being selected on the basis of specific speech characteristics without evidence of other problems (intelligence or hearing), demonstrated additional problems in other cognitive functions. Four cognitive functions were investigated: motor functioning, memory, sensory functioning, and attention. The results showed that the children with DAS had overall lower scores than the normally speaking children, and both groups showed a significant improvement at the second measurement more than one year later. Looking in more detail to the results of the children with DAS it was found that sequential abilities, both motoric and auditory, and sequential memory were significantly correlated with several speech characteristics of DAS (Maximum Repetition Rate Task and number of consonant substitutions and substitution in place of articulation). Furthermore, comparison of the scores of children with DAS on the second measuring moment with the scores of normally speaking children on the first measuring moment revealed an *overall delay* in development and an additional *deviance* in sequential abilities. The delay in development was interpreted as a concomitant effect of the disorder, that is, the speech deficit competes with other functions so that this may consequently delay the development of these other functions. A deviant development of cognitive functioning in DAS was especially found in the sequential abilities, concerning both sequential motor and memory tasks. When relating the general deviance in sequential abilities to the speech production model, the results of this study could give another suggestion for problems in phonetic planning and motor programming in children with DAS.

Finally, in the discussion we discussed the results of the experiments from a more global point of view, trying to answer the question whether there is one underlying deficit in DAS. An overview of the results made us conclude that both phonetic planning and motor programming of the speech production process are deviant in DAS. As part of the planning deficit, also deficits in 'higher' mechanisms, concerning durational control processes and use of prosody, seem to be at play in DAS. As part of the programming deficit, we also found evidence for problems in automation of speech production. From a neuropsychological point of view, these speech phonetic planning, motor programming, and automation deficits appeared to be embedded in more general information processing deficits, such as deficits in sequencing abilities and sequential memory. The latter results suggested a more generalized problem in sequencing and timing abilities.

We used a metaphor to illustrate this idea of problems in sequencing and timing. Imagine the speech production process to be an orchestra and a director performing a composition (assuming

that each member of the orchestra uses the same musical notation and is able to read this notation, as a metaphor for a correct phonological plan). If the instruments are not played in the correct sequence the music piece will sound awkward. But also the timing by which the successive instrument are played, requires conscientiousness. If not, some instruments may follow the others too quick, whereas others may be delayed. This will then result in inconsistent tempos and the composition will sound distorted. The performance of the composition by the orchestra stands or falls with the competence of the conductor to point to the successive instrument players in the right sequence and with the right timing. As in the orchestra, also in speech production sequencing and timing are very important. There are different 'instruments' involved in the production (e.g., the articulators and vocal folds) and the movements of the organs must be planned and programmed in the right sequence and the right timing. The speech characteristics of DAS as well as results of the experiments presented in this thesis suggest a generalized problem in sequencing and timing in DAS.

Samenvatting (summary in Dutch)

"Spraaikontwikkelingsdyspraxie: stoornissen in fonetische planning en motorische programmering"

Spreeken lijkt erg simpel voor een ieder die geen problemen ondervindt tijdens het spreken. Zij die ooit gewerkt hebben met kinderen of volwassenen met een taal- of spraakstoornis weten echter dat het vermogen om spraak te produceren niet altijd voor lief genomen kan worden. Dit proefschrift gaat over kinderen met een spraakontwikkelingsstoornis, namelijk spraakontwikkelingsdyspraxie (SOD) voor wie het produceren van verstaanbare spraak erg moeilijk is. Deze kinderen maken veel onregelmatige verwisselingen en vervormingen van spraakklanken waardoor bijvoorbeeld een woord als 'boek' de ene keer kan worden uitgesproken als 'koek', maar een volgend keer als 'toek' of 'broep'. Dit maakt dat de spraak van deze kinderen erg onverstaanbaar is. Hoewel de stoornis geschaard wordt onder de neurogene spraakstoornissen (dat zijn spraakstoornissen met een neurologische basis) zijn er tot op heden nog geen duidelijke aanwijzingen voor een neurologische aandoening bij kinderen met SOD. Evenmin is er een eenduidig antwoord gevonden op de vraag waar het probleem zich bevindt in het spraakproductieproces. Het ultieme doel van het onderzoek dat in dit proefschrift beschreven is was dan ook deze vraag te beantwoorden.

Als basis hiervoor hebben we een model van spraakproductie genomen dat ervan uitgaat dat een aantal opeenvolgende stappen doorlopen moet worden voordat men tot spreken komt. Dit spraakproductiemodel is uitgebreid beschreven in hoofdstuk 1 en wordt hieronder ook in iets meer detail toegelicht. Uitgaande van dit model is gekeken naar welke stappen een rol zouden kunnen spelen in het onderliggende probleem van SOD. De stappen die mogelijk verstoord zouden zijn in SOD zijn afzonderlijk onderzocht in verschillende experimenten. Deze experimenten zijn beschreven in de hoofdstukken 2 tot 6. Een samenvatting van de resultaten van deze experimenten volgt nadat eerst kort het spraakproductiemodel is uitgelegd.

Spraakproductiemodel: Welke stappen worden onderscheiden en welke zijn mogelijk verstoord bij SOD?

Een model biedt ons de mogelijkheid om een ingewikkeld proces, zoals spreken, makkelijker te begrijpen door het proces op te splitsen in relevante deelprocessen. In dit onderzoek zijn we uitgegaan van het model van taal/spraak-productie van Levelt (1989), waarin het proces van spreken is opgedeeld in drie deelprocessen, namelijk *conceptualiseren*, *formulator* en *articulator*. De boodschap van wat men wil zeggen wordt gevormd tijdens het conceptualiseren; hierin speelt kennis van de wereld een rol evenals kennis van conversatie (een leeftijdsgenoot wordt anders aangesproken dan een hoogleraar). Deze boodschap wordt doorgestuurd naar de formulator waar twee processen plaatsvinden. Ten eerste worden de lemma's (woordbetekenissen) bij de boodschap gezocht vanuit een mentaal lexicon (woordenboek) en daarmee worden grammaticale coderingsprocessen geactiveerd om de grammaticale of syntactische structuur te maken. Dit stelt ons bijvoorbeeld in staat om, wanneer we een vraag willen stellen, een andere volgorde van woorden te gebruiken dan wanneer we een mededeling doen, en om het werkwoord anders te

vervoegen indien er sprake is van één persoon of meerdere personen. Ten tweede wordt de syntactische structuur ingevuld met 'lexemen' (fonologische woordvormen) die ook vanuit het lexicon worden opgehaald en waarmee de fonologische codering wordt geactiveerd. Tijdens de fonologische codering worden de metrische structuur (dat is, het aantal lettergrepen, het klemtoonpatroon) en de segmenten (de klanken van een woord/frase) bepaald en samengevoegd (de klanken worden op de goede plaats in het woord-frame gezet). Het ontstane *fonologische woord* wordt doorgestuurd naar de articulator, om uitgesproken te worden. In ons onderzoek waren wij met name geïnteresseerd in de processen vanaf het fonologische woord. Weliswaar maken kinderen met SOD veel spreekfouten (onder andere door klankvervangingen), maar het soort fouten dat ze maken verschilt nauwelijks met dat van kinderen met fonologische spraakstoornissen en normaal sprekende kinderen. Dit lijkt er op te wijzen dat *het* probleem bij kinderen met SOD niet lijkt te liggen in de fonologische codering.

Dit eerste model van Levelt is later aangepast (Levelt 1999) en ook wij hebben nog een toevoeging gedaan vanuit een ander model (Van der Merwe, 1997) om de laatste stappen vanaf het fonologische woord tot het daadwerkelijk uitspreken nog wat te specificeren. De volgende stappen lijken hierbij belangrijk: *fonetische planning*, inclusief het gebruik van de *syllabary* (een soort lettergreepopslagplaats), *motorische programmering* en *motorische uitvoer* (zie figuur 1 in hoofdstuk 1). Tijdens de fonetische planning wordt het fonologische woord omgezet in een *fonetisch plan*, waarin de doelen van de articulatoren worden bepaald zowel ruimtelijk (bijv. tongpunt achter de tanden of lippen gesloten) als temporeel (bijv. een klinker 'o' duurt langer in 'koop' dan in 'kop'). Hiervoor zou gebruik gemaakt kunnen worden van vast opgeslagen patronen voor lettergrepen in de syllabary. Het fonetisch plan wordt vervolgens doorgestuurd naar de motorische programmering waar de exacte spraakbewegingen worden vastgelegd in een *motorisch programma*. Dit stelt ons in staat om toch verstaanbaar te spreken met een pen tussen de tanden; de doelen liggen dan vast in het fonetisch plan en tijdens de motorische programmering wordt de positie van de tong aangepast aangezien de kaak niet meer kan bewegen. Het motorische programma wordt vervolgens doorgestuurd naar de motorische uitvoer wat de aanstuur van de spieren regelt die bij spreken betrokken zijn.

Zoals hierboven al genoemd werd, waren wij voornamelijk geïnteresseerd in deze laatste drie processen in het onderzoek naar het onderliggende probleem bij kinderen met SOD. Deze processen hebben we onderzocht door kinderen bepaalde zinnen te laten herhalen en in hun uitingen (spraaksignalen) hebben we vervolgens akoestische metingen gedaan. Door middel van de akoestische metingen, namelijk spectrale formantmetingen en duurmetingen, is gekeken naar coarticulatie in de spraak. Coarticulatie is het proces dat bij uitspreken van een klank deze klank beïnvloed wordt door de omgevingsklanken, als gevolg van vooruit plannen (anticipatorische coarticulatie, bijv. de eerste 'k' in 'koek' laat al iets doorklinken van de 'oe') of voortdurende effecten (persevererende coarticulatie). Dit betekent dat voordat een klank uitgesproken wordt kenmerken van deze klank al invloed kunnen uitoefenen op voorafgaande klanken. Problemen in planning en/of programmering kan van invloed zijn op de samenhang van klanken en daarmee hun sporen nalaten in de coarticulatorische patronen. In dit onderzoek is de invloed van de klinker gemeten in de voorafgaande klanken door de tweede formant (F2) te bepalen; deze F2 is

kenmerkend in het onderscheid tussen de klinkers /u/ (spreek uit 'oe'), /a/ en /i/, namelijk een lage waarde in /u/ en een hoge waarde in /i/.

De experimenten en de resultaten.

Het eerste experiment, in hoofdstuk 2, was gebaseerd op een aspect van spraakontwikkeling dat ervan uitgaat dat in een normale ontwikkeling bij jonge kinderen meer coarticulatie (onderlinge afhankelijkheid en beïnvloeding van spraakklanken) gevonden wordt in de spraakuitingen dan bij oudere kinderen. Jonge kinderen beginnen met een meer globale articulatie die later in de ontwikkeling pas meer opsplitst in fonetische segmenten. Dit veronderstelt dat afzonderlijke fonetische segmenten pas aan het eind van de ontwikkeling ontstaan en dat de mate van coarticulatie iets kan zeggen over spraakontwikkeling(sstoornissen). In dit eerste experiment werden coarticulatie patronen van kinderen (normaalsprekend en kinderen met SOD) en volwassen vrouwen bestudeerd. Hen werd gevraagd om simpele betekenisloze uitingen te herhalen, zoals 'hé de ba weer', of 'he de si weer'. De resultaten lieten zien dat de normaalsprekende kinderen meer coarticulatie tussen de lettergrepen hadden (de invloed van de klinker op de schwa, dat is de 'e' in 'de') dan volwassen vrouwen. Hiermee werd de aanname van dit experiment, namelijk dat coarticulatie minder wordt gedurende de ontwikkeling, bevestigd. Inderdaad kunnen we zeggen dat spraak van jonge kinderen meer coarticulatie heeft dan die van ouderen.

Bij het vergelijken van spraak van kinderen met SOD met die van normaalsprekende kinderen werd gevonden dat kinderen met SOD veel variabler zijn in herhaalde uitingen, dat ze over het algemeen minder onderscheid maken tussen klinkers, en dat ze andere coarticulatie patronen hebben, zoals sterkere coarticulatie binnen de lettergreep. Bovendien vonden we dat de kinderen met SOD afzonderlijk idiosyncratische (eigenaardige) patronen lieten zien. Het verschil in mate van coarticulatie werd geïnterpreteerd als een teken van *achterstand* in ontwikkeling (sterkere coarticulatie wijst op jongere spraak). De eigenaardige patronen van de afzonderlijke kinderen echter, deden ons besluiten dat de spraakontwikkeling van deze kinderen ook *afwijkend* is. Dit zou met name verklaard kunnen worden uit afwijkende automatisering van spraakproductie. De grotere variabiliteit bevestigt het idee van onvolwassen automatisering. Kortom, uit het eerste experiment kwam naar voren dat kinderen met SOD zowel in mate als in kwaliteit verschillen laten zien in coarticulatie in vergelijking met normaalsprekende kinderen, wat zowel een vertraagde als verstoorde spraakontwikkeling suggereert.

Hoofdstuk 3 beschrijft een experiment dat is uitgevoerd om te onderzoeken of de fonetische planning van lettergrepen bij kinderen met SOD misschien verstoord is. Om dit te onderzoeken werden uitingen genomen die wat betreft volgorde van klanken gelijk waren, maar verschilden in lettergreepstructuur: 'ze schiet' tegenover 'zus giet' (let op, het gaat hier om de klanken zoals ze uitgesproken worden en niet om de schrijfwijze; 'ze schiet' wordt dan zonder pauze hetzelfde uitgesproken als 'zus giet'). We verwachtten een sterk effect van de lettergreepstructuur op de coarticulatorische samenhang en duurpatronen binnen de lettergreep (de [s] zal meer beïnvloed worden door de [i] in 'ze schiet' dan in 'zus giet'). Dit effect werd echter niet gevonden in de formant metingen van de normaalsprekende kinderen, noch in die van de kinderen met SOD. De duurpatronen lieten wel een effect van lettergreepstructuur zien in de tweede lettergreep, maar

alleen bij normaalspreekende kinderen. Systematische duuraanpassingen werden gevonden in de klanken van de tweede lettergreep, namelijk de medeklinker /x/ en de klinker werden korter als de /s/ toegevoegd werd in de tweede lettergreep. De afwezigheid van dit effect in de duurpatronen van kinderen met SOD werd opgevat als een stoornis in fonetische planning, en wel een gebrek aan duuraanpassing van de klanken aan de metrische structuur van de lettergreep. Dit suggereert dat in het spraakproductieproces van SOD elke klank afzonderlijk verwerkt wordt, zonder rekening te houden of aan te passen aan de omgevingsklanken binnen de lettergreep.

Effecten van de lettergreepstructuur op de coarticulatorische samenhang en duurpatronen *tussen* de lettergrepen werden gevonden als gevolg van prosodische verschillen tussen de uitingen. In normale spraak worden klanken in onbeklemtoonde lettergrepen meer gecoarticuleerd en zijn korter dan dezelfde klanken in beklemtoonde lettergrepen. Dit effect werd ook gevonden in de uitingen van de normaalspreekende kinderen, de onbeklemtoonde schwa (de 'e' in 'ze') in 'ze schiet' liet meer coarticulatie effect zien van de komende klinker en was korter dan de meer beklemtoonde schwa in 'zus giet'. Dit effect werd niet gevonden in de spraak van de kinderen met SOD. Het afwezig zijn van een dergelijk prosodisch effect op de coarticulatie en segment duur van de schwa ondersteunt het idee van een probleem in fonetische planning. Dit suggereert dat kinderen met SOD prosodische eigenschappen niet op dezelfde manier verwerken als normaalspreekende kinderen.

Ook in dit experiment werd grote variabiliteit in herhaalde uitingen gevonden bij kinderen met SOD, wat duidt op minder geautomatiseerde spraakproductieprocessen, voornamelijk in motorische programmering. Bewijzen voor het bestaan noch het verstoorde gebruik van een lettergreepopslag van vaakgebruikte lettergrepen konden niet gevonden worden.

De variabiliteit in herhaalde uitingen, zoals al beschreven werd in hoofdstuk 2 en 3, suggereren dat kinderen met SOD problemen hebben met (het automatiseren van) de motorische programmering. In hoofdstuk 4 is een mogelijk probleem in de motorische programmering nader onderzocht, met behulp van een bijtblok (een blokje dat tussen de kiezen geklemd moest worden en waardoor de kaak niet meer kan bewegen tijdens het spreken). Hoewel de normaalspreekende kinderen niet in staat waren tijdens het spreken volledig te compenseren voor het bijtblok (de F2 waarden waren hoger in de bijtblok spraak dan in de normale spreekconditie) had het bijtblok geen effect op de mate van anticipatorische coarticulatie. In dit opzicht lieten de normaalspreekende kinderen dezelfde effecten zien als de volwassen vrouwen. De kinderen met SOD waren evenmin in staat te compenseren voor het bijtblok, maar bij deze kinderen zagen we lagere F2 waarden in de bijtblok conditie. Bovendien werden sterke effecten van het bijtblok gevonden in de klinkerkwaliteit, die, in tegenstelling tot onze verwachtingen, verbeterd was. Het lijkt dus alsof de kinderen met SOD geholpen werden door het bijtblok. Mogelijkerwijs speelt een beperking van het aantal vrijheidsgraden een rol in het vergemakkelijken van de articulatie doordat bepaalde articulatiebewegingen niet gecontroleerd hoeven te worden. Evenwel vonden we een toename in de variabiliteit van de herhaalde uitingen in de bijtblok conditie, die erop wijst dat ondanks de verbetering van de klinkerkwaliteit, de kinderen met SOD toch moeite hadden met de articulatie tijdens het spreken met een bijtblok. Deze resultaten werden opgevat als een duidelijk bewijs voor een verstoorde motorische programmering in SOD, als gevolg van een minder geautomatiseerd en gecontroleerd proces.

In hoofdstuk 5 werd de controle van duren ('durational control') onderzocht in kinderen met SOD, teneinde een antwoord te krijgen op de vraag of de langzame spreesnelheid van deze kinderen een symptoom is van compensatie of een direct gevolg van de stoornis zelf. Hiervoor werd gekeken naar duurpatronen, dat is de intrinsieke en relatieve duren van klanken als gevolg van beïnvloeding van de fonetische duurcontext. En deze werden vergeleken tussen kinderen met SOD en normaalspreekende kinderen in twee spreekcondities, een normale spreekconditie en een bijtblok conditie. De duren van klanken in de normale spreekconditie lieten een sterke contextuele afhankelijkheid zien in de uitingen van de normaalspreekende kinderen. Namelijk, verschillen in intrinsieke duren tussen ploffers en fricatieven (de /s/ en /x/ - spreek uit 'g' - zijn langer dan de /b/ en /d/), en tussen hoge en lage klinkers (/i/ is korter dan /a/ en /u/), werden gecompenseerd in de duren van de omringende klanken. Dit systematische effect van intrinsieke duurverschillen en contextuele afhankelijkheid van de duren werd niet gevonden in de uitingen van de kinderen met SOD. Dit (gebrek aan) resultaat en de hogere variabiliteit in duren suggereren dat de controle van duren bij SOD minder strak is.

Deze duur controle mechanismen, specifiek betreffende compensatie strategieën, werden nader onderzocht door de duurpatronen in een bijtblok conditie te vergelijken met een normale spreekconditie. Normaalspreekende kinderen hadden langere duren in de bijtblokconditie, maar het duurpatroon bleef hetzelfde (zowel de klinker als de medeklinker werden lineair verlengd). Normaalspreekende kinderen vertraagden hun spraak dus als compensatie voor het bijtblok, maar controleerden daarbij voor de intrinsieke duren en de contextuele afhankelijk. Bij kinderen met SOD was het een ander verhaal. Deze kinderen verlengde de duur van de consonant, maar de duur van de klinker werd verkort. Dit resultaat is in overeenstemming met de suggestie dat een afwijkende controle van duren onderliggend zou kunnen zijn aan de spraakproductie van deze kinderen. Bovendien ondersteunen deze resultaat de suggestie, zoals al geopperd in hoofdstuk 3, dat kinderen met SOD elke klank afzonderlijk verwerken zonder aanpassing aan de fonetische context. Een probleem in de controle mechanismen van duren zouden ook deze resultaten kunnen verklaren. Controle mechanismen van duren worden niet specifiek genoemd in het spraakproductiemodel dat wij hanteerden. Maar als we de resultaten van hoofdstuk 5 en hoofdstuk 3 combineren zouden we kunnen concluderen dat controle van duren relevant is in zowel de fonetische planning (segment duren zijn afhankelijk van lettergreepstructuur) als in de motorische programmering (lange duren als gevolg van het bijtblok maar stabiele duurpatronen). Kortom, een gebrek aan controle van duren bij SOD beïnvloedt fonetische planning als ook de motorische programmering.

Hoofdstuk 6 beschrijft de resultaten van verschillende niet-spraak taken die afgenomen waren om te onderzoeken of kinderen met SOD, ondanks het feit dat ze geselecteerd zijn op grond van hun specifieke spraakkenmerken zonder andere problemen (intelligentie of gehoor), ook bijkomende problemen zouden kunnen hebben in andere cognitieve functies. Vier cognitieve functies werden hiervoor onderzocht: motorische functies, geheugen, sensorische functies, en aandacht. De resultaten lieten zien dat kinderen met SOD over het algemeen lager scoorden dan de normaalspreekende kinderen (van dezelfde leeftijd!), en dat beide groepen hoger scoorden op de tweede meting meer dan een jaar later. Toen we gedetailleerder naar de resultaten keken zagen we dat de scores van de sequentiëringstaken (daar waar volgorde van belang is) significant

samenhingen met de scores op de spreektaken (maximale repetitie snelheid en aantal medeklinkervervangingen en vervangingen van plaats van articulatie). Bovendien vonden we, toen we de scores van de kinderen met SOD op de tweede meting vergeleken met de scores van de normaalspreekende kinderen op de eerste meting (dus kinderen met SOD werden vergeleken met jongere normaalspreekende kinderen), dat de kinderen met SOD een achterstand in ontwikkeling lieten zien en een bijkomend probleem in sequentiële vaardigheden. De achterstand in ontwikkeling kan geïnterpreteerd worden als een bijkomend effect van de stoornis, dat wil zeggen de stoornis concurreert met andere functies waardoor de ontwikkeling van die functies achter zal blijven. Een verstoorde ontwikkeling van cognitieve functies in SOD vonden we op de taken waar sequentiële vaardigheden van belang waren, het ging om zowel sequentiële motorische taken als geheugen taken. Wanneer we dit algemene probleem in sequentiële vaardigheden relateren aan het spraakproductiemodel, levert dit ons een extra suggestie op voor problemen in fonetische planning en motorische programmering bij kinderen met SOD.

Tot slot werden in het discussie hoofdstuk (hoofdstuk 7) de resultaten en conclusies van de experimenten nog eens beschouwd vanuit een meer algemene invalshoek, teneinde de vraag te kunnen beantwoorden wat het onderliggende probleem is van SOD. Dit deed ons concluderen dat zowel de fonetische planning als de motorische programmering verstoord zijn in het spraakproductieproces van kinderen met SOD. Deeluitmakend van de fonetische planning lijken problemen in processen als controle van duren en het gebruik van prosodie relevant te zijn in SOD. Als deel van een probleem in motorische programmering vonden we ook aanwijzingen voor problemen in de automatisering van spraakproductie. Vanuit een neuropsychologisch oogpunt lijken deze problemen op het gebied van de spraakplanning, programmering en automatisering verankerd te zijn in een meer algemeen informatieverwerkingsprobleem, zoals stoornissen in sequentiële vaardigheden en sequentieel geheugen. Dit laatste suggereert een meer algemeen probleem in sequentiëring en timing. Aan de hand van een metafoor kan dit geïllustreerd worden.

Stel je het proces van spreken voor als een orkest met dirigent die een bepaalde compositie willen uitvoeren (vooropgesteld dat elk lid van het orkest de juiste notatie voor zich heeft en deze ook kan lezen, als metafoor voor een correct fonologisch plan). Zowel de volgorde als de timing van de bespeelde instrumenten is van belang. Als de instrumenten niet in de juiste volgorde gespeeld worden zal het stuk vreemd klinken, en als de timing van de instrumenten incorrect is zal het stuk met onregelmatige tempi gespeeld worden (soms sneller, soms langzamer) waardoor het vervormd klinkt. De uitvoering van het orkest staat en valt met de bekwaamheid van de orkest-dirigent om de juiste instrumenten (volgorde/sequentiëring) op de juiste momenten (timing) te laten bespelen. Evenals in een orkest is ook in spraakproductie sequentiëring en timing van essentieel belang. Er zijn verschillende ‘instrumenten’ bij betrokken (zoals de articulatoren en de stembanden) en de bewegingen van de instrumenten moeten gepland en geprogrammeerd worden in de goede volgorde en met de juiste timing. De spraakkaracteristieken van SOD en de resultaten van dit onderzoek suggereren een algemeen probleem van sequentiëring en timing in SOD. Dit biedt aanknopingspunten voor de therapie van kinderen met SOD.

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Curriculum Vitae

Lian Nijland is geboren op 23 mei 1972 te Bodegraven als vierde in een gezin van zes kinderen. Na de lagere school doorlopen te hebben ging ze in 1984 naar het Chr. Lyceum-Havo te Gouda waar ze in 1990 haar VWO- β diploma haalde. Vervolgens is ze aan de Universiteit Utrecht begonnen met de studie Psychologie met de bedoeling na het behalen van de propedeuse over te stappen naar de studie Fonetiek aan de Letterenfaculteit van dezelfde universiteit. Een jaar later vond de overstap daadwerkelijk plaats alhoewel ze met een half been bij Psychologie is gebleven. Na zes jaar, waarbij een stage werd gedaan aan het Instituut voor Doven te St Michielsgestel en tevens veel vakken gevolgd werden bij Psychologie, studeerde ze in 1996 af als Foneticus op een onderzoek naar de zangstem, 'Hysterese in stemgeving'. Na een jaar gewerkt te hebben bij de vakgroep Linguïstiek op een project voor de implementatie van geluidsdemonstraties op internet begon Lian in september 1997 aan een project met de titel 'Fonologische encoding bij kinderen met verbale ontwikkelingsdyspraxie', dat nog voor anderhalf jaar door NWO gefinancierd werd. Dit project kon vervolgens met andere financiering voortgezet worden wat heeft geresulteerd in dit proefschrift.

In de periode dat ze bezig was met haar promotie-onderzoek heeft Lian ook een pilot-studie gedaan naar articulatorische coördinatie in kinderen met spraakstoornissen met behulp van de articulograaf. Sinds juni 2001 is ze deels werkzaam bij het UMC Utrecht/ Wilhelmina Kinderziekenhuis, afdeling stem-, spraak-, taal- en gehoorcentrum, waar ze onderzoek doet bij kinderen met auditieve verwerkingsproblemen. Deze beide 'neven'-activiteiten en de afronding van het proefschrift hebben een goed gevolg gekregen in een postdoc onderzoek (binnen het NWO-onderzoeksprogramma 'Auditory perception in healthy speakers and in speakers with acquired or developmental language impairments'), waarin Lian sinds november 2002 op zoek is naar de 'Perception-production coupling in children with speech output disorders as compared to normal development'.

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